

**Doctoral Thesis**

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

**Psychological factors associated with distress and wellbeing in dystonia**

Doctorate in Clinical Psychology

Lancaster University

Helen Gowling

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**Statement of Total Word Count**

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<b>Section</b>	<b>Main text</b>	<b>Appendices, including References, Figures and Tables</b>	<b>Total</b>
Abstract	297	-	297
Literature Review	7,991	9,317	17,308
Research Paper	7,900	7,645	15,545
Critical Appraisal	3,627	1,225	4,852
Ethics Section	5,683	1,678	7,361
Total	25,498	19,865	45,363

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## **Thesis Abstract**

This thesis examines the psychological factors involved in distress and wellbeing for people living with dystonia.

Section one reports on a systematic literature review of quantitative studies examining the relationship between psychological factors and anxiety and depression in dystonia. A systematic search using key words relating to dystonia and concepts of psychological distress was conducted on four academic databases. Findings suggest that a number of psychological factors were related to anxiety and depression in dystonia. These included body concept, coping, personality traits, self-esteem, psychosocial domains of quality of life, and somatic complaints. The limited number of papers suggests that psychological factors are an under researched area when understanding the distress experienced in dystonia; nonetheless, the findings support a biopsychosocial model of understanding. This has clinical implications for the psychological management of distress for those living with dystonia and future research should seek to expand on this knowledge.

Section two reports on an empirical study examining the role of coping strategies in the relationship between stigma and psychological wellbeing in people living with cervical dystonia. Individuals with cervical dystonia completed an online survey including measures of stigma, coping strategies, quality of life, psychological distress and wellbeing. A series of parallel mediation models were conducted to explore the relationships between stigma, coping strategies and distress and wellbeing. Findings indicated that maladaptive coping strategies mediated the relationship between stigma and psychological distress and wellbeing. Adaptive coping strategies did not mediate the relationship between stigma and psychological distress and wellbeing. These findings support the need for holistic psychological formulations for individuals with cervical dystonia who are also experiencing psychological distress.

Section three includes the critical appraisal which reflected on the process of conducting this project. It discusses key decision points and critically evaluates some of the decisions made.

### Declaration

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

Name: Helen Gowling

Signature:



Date: 21/09/2020

## **Acknowledgements**

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On a personal level I want to say thank you to my friends and family for their support, especially my wonderful children who have been so patient and understanding whilst I have been on this journey. Finally, I want to give a special thanks to my fiancé Matthew. Thank you for your unwavering belief in me, even when I doubted myself. You are my rock.

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**Section One: Literature Review**

**Psychological correlates of anxiety and depression in dystonia**

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Helen Gowling

Doctorate in Clinical Psychology

Division of Health Research, Lancaster University

Prepared for submission to *Psychology & Health* (see appendix 1-2)

**All correspondence should be sent to:**

Helen Gowling  
Doctorate in Clinical Psychology  
Lancaster University  
Furness College  
United Kingdom  
LA1 4GY

Email : [h.gowling@lancaster.ac.uk](mailto:h.gowling@lancaster.ac.uk)  
Tel: 01524 59297

### Abstract

**Purpose:** Individuals with dystonia often experience depression and anxiety at higher rates than the general population. There is debate over whether these are primary symptoms of dystonia or secondary in response to living with a chronic health condition. The aim of this paper was to review psychological correlates of anxiety and depression for those living with dystonia.

**Methods:** PsycINFO, MEDLINE, CINAHL, and Web of Science databases were systematically searched for eligible studies using keywords related to ‘dystonia’ and ‘anxiety/depression’.

Quantitative studies which analysed the relationship between psychological factors and anxiety or depression were included. Quality assessment was conducted on all eligible studies.

**Results:** 16 studies were included in this review. Psychological correlates of anxiety and depression examined were body concept, coping, personality traits, self-esteem, psychosocial domains of QoL, self-efficacy and somatic complaints. In addition, some factors seemed to interact with each other to influence the overall measures of depression and anxiety. Other factors included in singular studies were locus of control, social support, sexual wellbeing, substance abuse and psychological symptoms as measured by the SCL-90.

**Conclusion:** The literature reviewed revealed limited research had been done in this area. What had been examined highlighted a number of different factors relating to distress for those living with dystonia. This supports a biopsychosocial understanding of the development of anxiety and depression. This has clinical implications for the psychological management of distress for individuals. Future research should explore the relationships between these factors using longitudinal designs and more complex statistical models.

*Keywords:* dystonia; anxiety; depression; movement disorders; psychology

## Introduction

Dystonia is a movement disorder characterised by involuntary intermittent or sustained contraction of muscles causing abnormal postures, tremors and twisting. These are often initiated or made worse by voluntary action (1). It is the third most prevalent movement disorder after Parkinson's disease and essential tremor (2) and the presentations of dystonia are wide ranging and varied (3). It has been proposed that dystonia be classified on two axes (4). Axis I specifies clinical characteristics such as age of onset, body distribution (focal, segmental, multifocal, generalised, hemidystonia), temporal pattern (disease course and variability) and associated features. Axis II specifies aetiology, stating whether the dystonia is inherited or acquired, idiopathic and any evidence of central nervous system pathology.

Due to the varied expression of the disease it is difficult to accurately assess current prevalence. Published epidemiological studies have estimated 15-30 cases of primary dystonia per 100,000 of the population (5,6), although this is likely to be an underestimation (1) as dystonia is often misdiagnosed or unrecognised (7). Indeed, a study investigating the prevalence of dystonia in the general community reported that it may be as high as 732 cases per 100,000 of the population, with only a small proportion of those identified having received an official diagnosis prior to the study taking place (8). Adult onset focal dystonias are the most common presentations (1) and within this category the most frequent types are cervical dystonia (CD, affecting the muscles of the neck and shoulders) and blepharospasm (affecting the eyes). Other presentations such as spasmodic dysphonia (SD, affecting the larynx) and writer's or musician's cramp (affecting the hand) are rarer (3).

People living with dystonia have been shown to experience high rates of psychological distress including depression, anxiety and social anxiety (1,9), with prevalence rates of between

12%-71% (10–15) compared to between 10%-20% in the general population of the UK (16). A recent study examined if type of dystonia had an impact on distress experienced (17). They found no difference between groups on measures of depression, but those with CD and SD scored higher than other focal types of dystonia on measures of anxiety. Additionally, those with SD scored higher than the other groups on measures of social anxiety.

The pathophysiology of dystonia is still not fully understood (3) and the underlying reason for the rates of psychological distress is under debate. There is evidence that, for some, psychological distress precedes the onset of dystonia motor symptoms (18–20) with this occurring in as many as 69% of participants in one sample (12). There have also been reductions in psychological distress following successful treatment with botulinum toxin injections or deep brain stimulation (21–23), as well as findings suggesting distress is related to disease severity (17,24). Additionally, longitudinal research has highlighted that those who have an improvement in the severity of their dystonia were less depressed compared to those whose dystonia had worsened (25), suggesting that depression and anxiety may be primary symptoms of dystonia with a biological basis. However, other longitudinal research suggests that distress does not improve in line with any improvement in the severity of dystonia (26) and that there is no relationship between dystonia severity and psychological distress experienced (27). Thus, there may be a secondary role for psychological distress, implying a complex spectrum of neurological and psychological symptoms (28).

Theories of dystonia have developed and expanded to include findings related to the neurological, motor and psychological features (29), along with insights from neurophysiological studies (2). As shown, there is conflicting evidence as to whether depression and anxiety are a primary feature of the disease or secondary to the impact of living with a movement disorder

(11,30). There is evidence to suggest that it could be a combination of both, with different factors interacting to contribute to the aetiology of depression, including a dysregulation of the metabolism of dopamine and monoamine, a genetic predisposition and adverse life events (31). Therefore, a biopsychosocial approach to understanding an individual's experience and difficulties would be beneficial. In addition, research in Parkinson's disease, has suggested that there may be three possible subtypes of depression in this population: those who would have been depressed if they had another chronic condition, those who would have been depressed without any condition, and those for whom the depression is related specifically to the underlying pathophysiology of the disease (32). This may also be the case in dystonia and could explain some of the contradictory findings within the research.

Despite the underlying mechanisms of anxiety and depression in dystonia remaining unclear, both have been shown to reduce the quality of life for those who experience them (9,10,33,34), as well as contributing to overall levels of disability (11,13,15,35). Given that the research on distress in dystonia has tended to focus on the medical model to date, it would be pertinent to give attention to the psychological factors within the biopsychosocial framework of understanding (31). This would act as an impetus for future research as well as gaining an understanding of psychological factors that may be related to anxiety and depression. This understanding would enable formulation-based interventions to help those who were experiencing psychological distress in addition to dystonia. A psychological formulation is a collaboratively constructed hypothesis about a person's difficulties which takes into account a range of factors including social circumstances, relationships and life events (including the presence of disease) and uses this understanding to plan how to move forward (36).

For the purposes of this review the term ‘psychological factors’ is used to describe factors that are potentially modifiable through psychological intervention, and is in line with how it has been defined in previous comprehensive reviews in other movement disorders (37,38).

Therefore, this review of the literature aims to examine:

- a) Which psychological factors have been investigated in dystonia?
- b) What is the relationship between these factors and anxiety and depression?

### **Method**

The framework adopted for the reporting of this systematic review was the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (39).

#### ***Search strategy***

A systematic search of four databases was conducted: CINAHL, MEDLINE, PsycINFO and Web of Science. These databases were chosen following guidance from an academic librarian and because they covered both medical and psychological research, thus increasing the chances of finding relevant papers on dystonia. These were also similar databases to those used in a previous review in dystonia (40), with the addition of Web of Science as it is recommended as an optimal database for efficient coverage (41). The final search was performed on November 25<sup>th</sup>, 2019. Database thesauruses and Medical Search History (MeSH) terms were searched for key words relating to dystonia and anxiety/depression. The search strategies also included free-text searches to capture any articles that may not be indexed correctly. Full details of the search strings are available in table 1-1. Searches were filtered to only include peer-reviewed literature in the English language. Once the search had been conducted, duplicates were removed, and the remaining papers screened for suitability.

[Table 1-1 about here]

***Inclusion criteria***

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

- Were quantitative empirical research.
- Participants were adults (aged >16 years) who had a diagnosis of primary dystonia.
- Included a validated measure of anxiety or depression, or diagnosis via gold standard diagnostic interview.
- Included a psychological factor (e.g. coping styles, personality traits).
- Reported a relationship between the psychological variable and anxiety or depression.

Studies were included if psychological factors were looked at in relation to anxiety or depression even if this was not the primary aim of the study. The subscales of quality of life measures that met this criterion (i.e. those that pertained to social and emotional role functioning) were included. The ‘validated’ measures of anxiety and depression had been established within research as having reliability and validity. Papers were excluded if dystonia was secondary to another neurological condition or as an extrapyramidal effect of medication.

***Data extraction and quality assessment***

Data extraction was performed by the author. Information extracted from each study included the sample size, age and sex of participants where stated, design and analysis, the country where the study was conducted, the psychological variables examined, and measures used to assess depression and anxiety. Information was also extracted to assess the risk of bias (study quality).

The quality of selected studies was assessed using the Appraisal tool for Cross-Sectional Studies (AXIS tool) (42). This scale is designed for cross-sectional research and consists of 20 questions that measure aspects of study quality including clear aims and objectives, the representativeness of the sample, information regarding non-responders, whether validated measures were used, statistical methodology and any conflicts of interest (see appendix 1-1 for full list of questions). To score the studies, each question was rated on a binary scale with 1 for questions that could be answered 'yes' and 0 for questions where the answer was 'no' or 'not determined'. There were two exceptions to this; questions 13 and 19 where the phrasing meant that an answer of 'yes' would suggest a higher risk of bias, so these were reverse scored. Following the assessment each study was given a score between 0-20, with higher scores representing a lower risk of bias and higher quality of research. A colleague also performed a quality assessment of a random sample of eight (50%) of the papers to check for bias, followed by a discussion to resolve a few minor discrepancies which were related to the interpretation of a couple of the questions. For example, the question 'Were the results for the analyses described in the methods, presented?' was interpreted by one colleague to mean the analyses should be in the methods section. The other colleague gave a score if the analyses were described in the methods or results section. Final scores were agreed on by both parties.

## **Results**

### ***Study selection***

The initial literature search returned 2148 articles from the databases CINAHL (n=97), PsycINFO (n=554), Medline (n=262) and Web of science (n=1235). Duplicates were removed leaving 1814 articles for screening. The titles and abstracts of these articles were examined according to the inclusion and exclusion criteria leaving 57 articles for a full text examination.

The reference lists of these articles were also screened for any further studies, but none were identified. Following full text examination, 41 studies were excluded due to not including anxiety/depression as correlates (n=21), not using a validated measure (n=2), a relationship between anxiety depression and other psychological factors not being examined (n=15) and using the same cohort and analysis as another study (n=3). This left 16 studies which were eligible for inclusion in this review. A flow diagram of the selection process can be seen in figure 1-1.

[Figure 1-1 about here]

### ***General study characteristics and quality appraisal***

Table 1-2 shows a summary of the studies included in the review. All the studies included used a cross-sectional design. Correlational analyses were used in seven papers and five carried out multiple regressions. Three papers used between groups analyses and one used a cluster analysis. Four of the studies were carried out in the UK (24,43–45), four in Germany (27,28,46,47), four in the USA (48–51) and one each in Austria (52), Serbia (53), Italy (54) and The Netherlands (55). Focal dystonia was examined in 14 of the studies (27,28,43,44,46–55) with the remaining two (24,45) using a mixed sample. Two studies also included patients with hemifacial spasm within their participants (24,54), though, as they were a small percentage of a large sample, the decision was made to include the findings in this review.

Recruitment for the studies was primarily via hospitals or clinics (27,28,46,47,49,50,52–55). Three recruited using an existing epidemiological sample (24,45,51), although two of these were from the same epidemiological sample (24,45). One study recruited from a mailing list of people previously involved in research (44) and another study used a subgroup of these participants (43). One study did not specify from where the sample was recruited (48).

The studies included represented a total of 1585 participants, not double counting the studies which used the same pool of participants (24,43–45,49,50). For those studies which included male and female participants and stated the percentage of each gender (n=13), the number of males ranged from 25.4% to 80% with a median of 40.8%. For the studies which indicated ages (n=12) these ranged from 17-91 years with mean ages ranging from 40.9-64 years. For studies that mentioned disease duration (n = 7) this ranged from 6 months to 56.6 years. For those that included mean age of onset (n = 9) this ranged from 34.7 years to 56.3 years.

[Table 1-2 about here]

### *Quality appraisal*

Table 1-3 outlines the quality appraisal using the AXIS tool (42). The scores from the quality appraisal ranged from 12 to 20 out of a possible 20. The main issues which contributed to lower scores were not justifying sample sizes and not addressing non-responders. Sample sizes ranged from 18 to 329, with a median of 86. One study justified their sample size with a sample size calculation for a power of 0.85 (55). Participant characteristics were described in all of the studies and 11 addressed non responders (24,27,28,43–45,47,49–51,55). Response rates were stated in six studies and ranged from 68% (44) to 92.4% (27). For the 68% response rate it was reported that there were no differences between the responders and non-responders in terms of clinical and demographic variables (44).

Three studies took steps to account for multiple statistical tests. One utilised the Bonferroni correction (46) and two adjusted their significance level to  $p < 0.001$  (43) and  $p < 0.005$  (45). Other studies that used a high number of statistical tests without taking steps to mitigate this were more vulnerable to type 1 error (24,28,44,47,48,50,51,53–55).

[Table 1-3 about here]

### ***Outcome measures***

The majority of the studies looked at both anxiety and depression (27,28,47–51,53–55). Five looked at only depression (24,43–45,52) and one at only anxiety (46).

A number of different measures were used to assess depression and anxiety. The following measures were used to assess depression: the Beck's Depression Inventory (BDI) (56) (n=7) (24,43–45,52,54,55), the Beck's Depression Inventory II (BDI-II) (57) (n=1) (51), the Self-Rating Depression Scale (SDS) (58) (n=1) (48), the Patient Health Questionnaire-9 (PHQ-9) (59) (n=1) (51), and the Hamilton Depression Rating Scale (HDRS) (60) (n=1) (53).

Anxiety was examined using the following measures: the State-Trait Anxiety Inventory (STAI) (61) (n=2) (48,54), the Hamilton Anxiety Rating Scale (HARS) (62) (n=1) (53), the Competitive Trait Anxiety Inventory (CTAI) (63) (n=1) (46), the Social Phobia Scale (SPS) (64) (n=1) (27), the Beck Anxiety Inventory (BAI) (65) (n=1) (55) and The Liebowitz Social Anxiety Scale (LSAS) (66) (n=1) (51). The following measures looked at both depression and anxiety: the Hospital Anxiety and Depression Scale (HADS) (67) (n=3) (49–51) and the anxiety and depression subscales of the Symptom Checklist-90 (SCL-90) (68) (n=1) (47).

Two studies (27,28) assessed for psychiatric comorbidity (including anxiety and depression) using the Structured Clinical Interview for DSM-IV (SCID) (69).

### ***Prevalence of anxiety and depression in the samples***

Current prevalence of anxiety and/or depression was reported in nine of the studies (24,27,43,44,48,50–52,55). The prevalence rates for depression ranged from 2.8% (50) to 53.4% (27). The prevalence rates for anxiety ranged from 11% for state anxiety (48) or 13.4 % for general anxiety (50) to 50% (27). Hu (50) commented on the particularly low rates of anxiety

and depression in their sample compared to the wider literature, suggesting that their participants had been living for a long time with SD (mean duration >10years) and were established on effective doses of botulinum toxin. In addition, the rates for anxiety in the study conducted by Gundel et al.(27) were higher than has been reported in previous research. This may be because they modified the social phobia diagnosis in the DSM-IV to allow it to be applied to those with a disabling or disfiguring physical condition. Traditionally these participants would not have met the criteria for this diagnosis, therefore increasing the prevalence rates in this study.

### *Correlates of anxiety and depression*

#### *Body concept*

Body concept (BC) is the conceptual image about one's own body and was examined in five studies in relation to anxiety and depression (24,27,43–45). Four of these (24,43–45) used the 'Body Concept Scale' developed by Jahanshahi and Marsden (25), where a higher score indicates a more negative BC. Two studies also included a measure of 'disfigurement' (24,44), a single item rating scale designed to assess the degree of self-perceived disfigurement. Jahanshahi and Marsden (44) found that BC was significantly correlated with depression in CD with a large effect size, with a more negative BC relating to an increase in depressive symptoms. However, when entered into a hierarchical multiple regression analysis alongside disfigurement, extraversion, neuroticism, and functional disability, BC accounted for less than 1% of the variance of depression. They suggested that the contribution of BC was minimal once the effects of perceived disfigurement had been accounted for, with disfigurement accounting for 13.5% of the variance in depression. Jahanshahi (43) again found that a more negative BC was associated with an increase in depressive symptoms in CD (with a large effect size). However, it was not a significant predictor in a stepwise multiple regression when included alongside self-depreciation,

disability, maladaptive coping, satisfaction with social support, belief in control by powerful others and three ratings of clinical severity. Lewis et al. (24) extended these studies to include participants with focal, generalised or segmental dystonia. They found that negative BC was still associated with an increase in depressive symptoms. In a stepwise multiple regression alongside self-esteem, disfigurement and quality of life, BC accounted for 6% and disfigurement accounted for 2% of the variance of depression scores. This variance accounted for by BC was maintained once the analyses was repeated to exclude participants with CD suggesting that this relationship affects those with other types of dystonia. However, disfigurement was no longer a significant variable once the analyses was repeated to exclude participants with CD suggesting this concept may be more relevant to those with CD than other types of dystonia. Page et al. (45) also included participants with varying types of dystonia. They looked at depression as a predictor of BC and found that scores on the BDI accounted for 43% of the variance in BC, highlighting the potential bidirectional relationship between these two concepts.

In the Gundel et al. (27) study, participants assessed their body image dissatisfaction on a self-rating visual analogue scale (VAS), with higher values indicating greater body image dissatisfaction. In their stepwise multiple regression analysis, they found that body image dissatisfaction was one of the independent predictors of social phobia when examined alongside education, marital status, tsui score (severity of CD), extent of CD, frequency of Botox, pain, significant life event in year prior to diagnosis and depressive coping.

### *Self-esteem*

Self-esteem was examined in two studies (24,43). In Jahanshahi (43) this was divided into self-worth and self-depreciation using the Rosenberg's self-esteem scale (RSES) (57). Their findings showed that the participants who were moderately to severely depressed, as measured

using a cut off score of 17 on the BDI, had significantly lower self-worth and significantly higher levels of self-depreciation. Both of these measures were also strongly correlated with depression with a large effect size. In a stepwise regression analysis alongside disability, degree of head control, satisfaction with social support and maladaptive coping; self-depreciation was the strongest predictor of depression, accounting for 59% of the variance. A further regression analysis with self-depreciation as the outcome variable showed that BC and maladaptive coping accounted for 52% and 7% respectively of the variance in self-depreciation.

Lewis et al. (24) also used the RSES but used an overall score of self-esteem. They found that self-esteem was negatively correlated with depression with a medium effect size and, in a stepwise regression analysis, accounted for 56% of the depression scores when controlling for BC, disfigurement and quality of life. This only dropped slightly to 55% once they repeated the analysis to exclude participants with CD. This study also looked to isolate the contributors to self-esteem, and again found that BC was the strongest predictor, explaining 38% of the variance in self-esteem.

### *Coping*

Four studies looked at coping styles in relation to psychological distress (27,43,46,47). The 'Freiburger Fragebogen zur Krankheitsverarbeitung' (FKV) (58) was used by two of the studies as a tool to examine coping strategies in people living with CD (27,47). Scheidt et al. (47) found that higher scores in depressive coping (described as cognitions and emotional attitudes towards the disease such as feeling guilty and blaming oneself), active problem focussed coping, and wishful thinking (described as denial and escapism into fantasy) were all related to increases in anxiety and depression. The effect size of wishful thinking and active problem focussed coping was small and the effect size for depressive coping was medium.

Gundel et al. (27) found that, in a stepwise regression alongside body image dissatisfaction, clinical variables and demographic variables, depressive coping was the main predictor of psychiatric comorbidity (including anxiety and depression) and social phobia.

Ioannou and Altenmueller (46) used the Stress Coping Questionnaire (SVF-78) (59) and found that coping strategies of ‘mental perseveration’ (described as being unable to break off from thoughts) and ‘self-incrimination’ (described as attributing stress to one’s own mistakes) were correlated with the self-doubt concern and somatic anxiety subscales of the CTAI with a medium to large effect size in focal hand dystonia. They also found that the coping strategy of ‘flight tendency’ (described as using thoughts of escape) was not significantly correlated with the subscales of the CTAI. In Jahanshahi’s (43) study the ‘ways of coping’ checklist (73) was used, adapted for chronically ill populations (74). They divided the coping strategies into either ‘maladaptive coping’ for those that were associated with increased depressive symptoms (wish fulfilling fantasy  $r = 0.48$ , religious faith  $r = 0.24$ ), or ‘adaptive coping’ for those associated with fewer depressive symptoms (threat minimisation  $r = -0.46$ , positive appraisal  $r = -0.08$ , cognitive restructuring  $r = -0.18$ , instrumental coping  $r = -0.04$ ). When entered into a stepwise multiple regression alongside self-depreciation, disability, degree of head control, and satisfaction with social support, maladaptive coping explained 1.7% of the variance in depressive symptoms.

### ***Personality traits***

Personality traits were examined in three studies (28,44,46). Specifically perfectionism (46), extraversion (28,44), neuroticism (28,44) and openness (28). Ioannou & Altenmueller (46) examined perfectionism in musicians with focal hand dystonia using the Frost Multidimensional Perfectionism Scale (FMPS) (62). They found that ‘concern over mistakes’ and ‘doubts about

actions' were both positively correlated with the 'self-doubt concern' and 'somatic anxiety' subscales of the CTAI with medium to large effect sizes.

Extraversion and neuroticism were examined in two studies (28,44). Jahanshahi & Marsden (44) measured these using the Eysenck Personality Questionnaire (76) and Lencer et al. (28) used the NEO five factor inventory (NEO-FFI) (64). Jahanshahi & Marsden (44) found that depression was positively correlated with neuroticism and negatively correlated with extraversion, both having medium effect sizes. They also found that, when entered into a stepwise regression alongside functional disability, BC and disfigurement, extraversion accounted for 14% and neuroticism accounted for 13% of the variance of depression. Lencer et al. (28) looked at the difference between groups of those with primary focal dystonia either affected by, or not affected by, a psychiatric disorder as measured using the SCID. They found the participants who met the criteria for a diagnosis of major depression or anxiety scored significantly higher on scores of neuroticism than those who did not meet the criteria. This difference was not observed on scores of extraversion. Those who met the criteria for social phobia scored significantly higher on the scale of neuroticism, and significantly lower on the scale of extraversion than those without social phobia. They also looked at scores of 'openness' and found no significant differences between those with anxiety, depression or social phobia and those without. Despite the different findings, both studies examining extraversion were similar in terms of quality which was rated as good.

### ***Self-efficacy and locus of control***

Self-efficacy was examined in two studies (49,50) relating to anxiety and depression in participants with SD. Both studies used the same sample of participants but different analyses. Self-efficacy was measured using the General Self-Efficacy Scale (GSES) (78) alongside a

Disease Specific Self-Efficacy in SD Scale (DSSES) which was developed by the authors in the first paper and based in part on the GSES (49). In Hu et al. (49) they found that higher levels of general self-efficacy (GSE) were significantly correlated with lower levels of anxiety and depression with medium effect sizes, and that higher levels of disease specific self-efficacy (DSSE) were significantly correlated with lower levels of anxiety and depression with medium to large effect sizes. Hu et al. (50) entered these variables into a linear regression model alongside demographic variables and measures of clinician and subjective ratings of voice. They found that GSE was a significant predictor of anxiety but not depression ( $p < 0.001$ ) and that DSSE was a significant predictor of depression but not anxiety ( $p < 0.001$ ). However, the linear regression models included both measures of self-efficacy which may have correlated highly with each other leading to issues of multicollinearity.

In the analysis done by Jahanshahi (43) locus of control was examined using the Multidimensional Health Locus of Control Scale (MHLOC) (79). This is divided into three subscales relating to belief in control by internal, chance and powerful others. Correlation analysis showed that a higher level of belief in internal control was significantly correlated with lower levels of depression with a small to medium effect size, whereas a higher belief in the control of powerful others was significantly correlated with an increase in depression with a large effect size.

### ***Psychosocial Subscales of Quality of Life Measures***

Two studies looked at depression/anxiety in relation to the psychosocial subscales of the SF-36 quality of life measure (53,55). In a univariate analysis Smit et al. (55) found that lower scores on the domain of 'social functioning' were significantly correlated with increased depression with a medium effect size, but not anxiety (though this had a small effect size). They

also found that lower scores on ‘emotional role limitation’ were significantly correlated with an increase in both anxiety and depression in participants with CD, with a medium to large effect size.

Pekmezovic et al. (53) included participants with CD, blepharospasm and writer’s cramp. Lower scores in social functioning were significantly correlated with higher levels of depression for all groups (large effect sizes) and with anxiety for CD and blepharospasm (medium to large effect sizes) but not writer’s cramp despite this still having a medium effect size. Lower scores on the emotional role limitation domain were significantly correlated with an increase in anxiety and depression for CD and blepharospasm (medium to large effect sizes) but not writer’s cramp despite this also having a medium effect size for both anxiety and depression. Out of these two studies, Smit et al (55) was of a higher quality.

### ***Social support***

Only one study considered the role of social support in relation to depression in CD (43) which was measured using the Social Support Questionnaire (80). This measure looks at both the amount of social support available and the individual’s satisfaction with that support. Correlation analyses found that a lower amount of support, and low levels of satisfaction with support were significantly correlated with an increase in depressive symptoms with a medium effect size. In a stepwise multiple regression, satisfaction with social support accounted for 1.6% of the variance in depression scores when entered into the model with self-depreciation, disability, degree of head control and maladaptive coping.

### ***Sexual wellbeing***

Sexual wellbeing was considered in one study looking at CD and blepharospasm (54). This was evaluated using the Sexual Functioning Inventory (SFI) (81) which has the subscales of

infrequency, non-communication, dissatisfaction, avoidance, non-sensuality, premature ejaculation/vaginismus and impotence/anorgasmia. Their analysis showed that a lower level of sexual wellbeing (as indicated by a higher overall score on the scale) and high scores of infrequency were significantly correlated with an increase in depressive symptoms with a small to medium effect size. High scores of sexual infrequency were also significantly correlated with an increase in anxiety with a medium effect size.

### ***Substance Abuse***

One study looked at the role of substance abuse in CD (51). Their analysis found that participants who met the criteria for substance abuse as determined by the SCID had a significantly higher level of depressive symptoms (as measured by the PHQ-9 and BDI-II) and distress (as measured by the HADS total score). As the HADS total score was used it is not clear whether this also suggested higher rates of anxiety. Social anxiety specifically as measured by the LSAS was not significantly different in those who met the criteria for substance abuse compared to those who did not.

### ***Other psychological concepts***

Other psychological concepts were examined in two of the studies, somatic complaints (48,52) interpersonal sensitivity, obsessive compulsiveness and psychoticism (52). Moraru et al. (52) used the subscales of the Symptom Checklist (SCL-90) (68) to examine whether there was any correlation with depression as measured by the BDI in participants with CD. The sample size in this study was small (n=40). The findings suggested that those who were moderately/severely depressed (n=5) scored significantly higher on the subscales of somatization, obsessive-compulsiveness symptoms, interpersonal sensitivity and psychoticism when compared to non-depressed participants (n=23). When they compared the non-depressed participants with those

who scored as mildly depressed (n=12) they found a significant difference in scores for obsessive-compulsiveness, hostility and somatisation.

Cannito (48) also examined somatic complaints in a sample with SD. The measure used was a Somatic Complaints Checklist developed by the author. They found that an increase in somatic complaints were significantly correlated with an increase in depression and state anxiety, both with a large effect size. There was no significant correlation between somatic complaints and trait anxiety although there was a medium effect size ( $r = 0.29$ ), so the lack of statistical significance may be due to the study being underpowered to detect this effect (n=18). Both studies examining somatic complaints were of a low quality.

### **Discussion**

This review sought to examine which psychological factors have been investigated in dystonia and what their relationship was with anxiety and depression. The limited number of studies identified (n=16) suggests that there has been little focus on psychological correlates of anxiety and depression in dystonia. The most frequently examined factors were BC, coping and personality traits followed by self-esteem, psychosocial domains of QoL, self-efficacy and somatic complaints. In addition, some of these factors seemed to interact with each other to influence the overall scores of depression and anxiety. Other factors were only included in single studies including locus of control, social support, sexual wellbeing, substance abuse and psychological symptoms as measured by the SCL-90.

BC was the most examined psychological factor in relation to depression. This is a multifaceted concept which incorporates an individual's feelings and beliefs about their body including an evaluation of how it is perceived by others (82). Negative BC has been shown to be experienced by people with dystonia and a review found it was related to poorer quality of life

outcomes (83). The results here suggest it is also related to depression with a large effect size. When examined alongside a variety of other demographic and psychological variables, the reliability of BC as a predictor of depression varied when perceived disfigurement was included as another variable. The results were not consistent between studies. Lewis et al. (24) suggested a larger role of BC, and Jahanshahi and Marsden (44) found that disfigurement accounted for the majority of the variance in depression. In addition, Lewis et al. (24) found that disfigurement was no longer a significant predictor when those with CD were removed from the analysis. This suggests there is some complexity and overlap between these two constructs which may warrant further investigation, as well as a difference between different types of dystonia. BC was found to be a reliable predictor of social phobia (27), although this study did not include a measure of perceived disfigurement. The findings also support a relationship between BC and self-depreciation which adds another layer of complexity into these potential relationships. Sociologists generally consider the body to be central to our self-identity and the image we have of ourselves (84). Research into body image has shown that there is a strong relationship between levels of self-depreciation and an individual's view of their own body image (85). Body image has also been looked at in relation to multiple sclerosis (86) showing evidence for the impact this has on psychological outcomes such as depression and self-esteem. Both self-esteem and self-depreciation were correlated with depression with large effect sizes in this review. In addition, one study looked specifically at whether depression predicted BC (45), highlighting the potential bi-directional relationship between these two concepts. Further longitudinal research could help identify in more detail how BC, disfigurement, and self-esteem may be interrelated and impact on each other as well as on the experience of psychological distress.

After BC, coping strategies were the next most examined concept. Coping is thought to mediate the relationship between stress, including that related to a chronic health condition, and psychological outcomes such as anxiety and depression (87,88). Coping strategies were found to correlate with anxiety and depression with small to large effect sizes and were independent predictors when entered into regression models. Coping styles were generally divided into 'adaptive' and 'maladaptive'. Maladaptive coping styles included depressive coping, wishful thinking, mental perseveration, self-incrimination, flight tendency and religious faith. Adaptive coping styles included 'active problem focussed' coping, threat minimisation, positive appraisal, cognitive restructuring and instrumental coping. The results from the review regarding coping were largely in keeping with the wider literature that suggests maladaptive coping styles can lead to increased levels of distress (89–91). One of the exceptions to this finding was that 'active problem focussed' coping, which would be classified as a more 'adaptive' strategy, was related to higher levels of anxiety in one of the studies, albeit with a small effect size (47). This may be because the impact of coping strategies can also be dependent on the specific context of the stressful situation (92,93). There is evidence that coping styles are not necessarily utilised in isolation (94). Therefore, participants may have used maladaptive coping strategies alongside the adaptive coping strategies, or utilised 'active problem focussed' coping in relation to a specific stressor. These findings suggest the relationship between distress and coping is complex and that it might be useful to consider the context in which a coping style is used before deciding if it is adaptive or not. In addition, the cross-sectional design of the study also limits how much can be inferred from the findings. For example, it may be that the anxiety had prompted the use of the active problem solving, rather than being a result of this, or indeed both the increase in anxiety and the increase in problem solving could be due to other underlying factors. Furthermore,

categorising coping in ways such as ‘maladaptive’ and ‘adaptive’, although useful for broader investigation, can undermine the potential relationships between strategies and the complexity of managing stress relating to chronic illness (92,95). Nonetheless there is some evidence to support the role that coping plays in psychological distress and attention should be given to this in future research.

Personality traits (neuroticism, extraversion and perfectionism) were looked at across a range of dystonia types and found to be related to anxiety and depression with a medium effect size. They were reliable predictors when examined alongside a variety of other demographic and psychological variables. Personality is thought to affect coping styles and emotional responses to the experience of having a chronic illness (96) and have been examined in the broader general literature on psychological distress and wellbeing (97,98).

Neuroticism has also been linked to individuals experiencing negative emotional responses in threatening situations (99) which could be related to the stress of living with a chronic condition. There were differences between two studies on whether extraversion was related to lower levels of depression (28,44) despite both studies being of a similar quality, but the null finding of Lencer et al. (28) may have been due to the small sample size (there were only five participants in the ‘depressed’ group) or the fact that Jahanshahi and Marsden (44) used the BDI whereas Lencer et al. (28) used the SCID. Perfectionism has been shown to lead to lower levels of wellbeing and has also been linked to higher levels of neuroticism (100) suggesting there may be interplay between different personality traits. In addition, the concept of extraversion has been shown to have different facets which may not have been captured in the measures used, and which have different relationships with depression (101).

Higher levels of self-efficacy, as measured by both a general and a disease specific scale, was found to be related to lower levels of distress with medium to large effect sizes. They were also reliable predictors of anxiety (GSE) and depression (DSSE) (49,50). Self-efficacy is an individual's belief that they are able to achieve a specific goal or task and has been shown to have a strong relationship with health behaviours (102). It has also been associated with psychological outcomes in other movement disorders such as multiple sclerosis (103). The findings in this review also considered the relationship between locus of control (the belief someone has in how much control they, or external forces, have over a situation) and psychological distress. Other areas of health research have suggested that high levels of a belief in chance, or in powerful others, can lead to increased levels of distress and lower quality of life (104,105) and these findings were echoed in this review. However, an external locus of control may also be seen as being more realistic and adaptive for conditions which are considered chronic (106). Locus of control has been shown to change over the course of an illness (107) which suggests that duration of disease may also be a relevant factor when examining this in the research. However, in the reviewed studies the disease duration of participants was too varied to draw any conclusions as to possible interactions between this and locus of control.

Psychosocial subscales of a quality of life measure, namely 'emotional role limitation' and 'social functioning', were found to be related to anxiety and depression across different types of dystonia with medium to large effect sizes. The SF-36 is a commonly used measure of quality of life and has been frequently used in dystonia (108). Concerns have been raised about its use with other neurological disorders (109–111) and an analysis of the measure in the context of dystonia found that the 'role emotional' scale had large floor and ceiling effects (112). This suggests this subscale may underestimate any clinical change and the results should be interpreted with

caution. However, the 'social functioning' subscale was found to be reliable and can be interpreted with more confidence (112). Although the two studies showed differing results in relation to the link between anxiety and social functioning, the study by Smit et al. (55) showed findings that approached significance and that had a small effect size. In addition, this study was rated as a higher quality than Pekmezovic et al. (53) which may add more strength to their findings.

The findings from Moraru et al. (52) suggested that somatization, obsessive-compulsive, interpersonal sensitivity, hostility and psychoticism all played a role in depression, and similar findings have been observed in Parkinson's disease (113). The findings that somatic complaints were correlated with anxiety and depression should be interpreted with caution as the scales used to examine anxiety and depression also include questions regarding somatic complaints. In research looking at the role of somatization in disability, the unique contributing effect of somatization reduced when anxiety and depression were added to the model (114) posing a challenge when formulating the relationships between these constructs.

In addition to the single studies examining locus of control and the SCL-90, sexual wellbeing, social support and substance abuse were also only investigated once. Aspects of sexual wellbeing were correlated with anxiety and depression with medium effect sizes. Social support was correlated with depression with a medium effect size and was also an independent predictor in a regression model. Substance abuse was found to be related to higher levels of depression and distress. As these were only looked at in singular studies it can be hard to draw conclusions beyond highlighting the need for conducting more research within this area.

### *Clinical Implications*

The range of psychological variables looked at in relation to anxiety and depression highlights the need for individual and holistic psychological formulations for people with dystonia who are also struggling with psychological distress. However, evidence for effective behavioural interventions in dystonia is limited (40). Treatment options tend to be based on recommendations developed for non-dystonic populations (35) and the recommended treatment for depression, anxiety and social phobia is cognitive behavioural therapy (CBT) (31,40,115). One study has examined the use of CBT and mindfulness in dystonia (115) and their findings suggested that a three-day residential programme utilising these approaches improved wellbeing and reduced distress. However, they caution that inferences from their data are limited due to the low number of participants (n=9). Qualitative data from the study also suggested that mindfulness was used as a coping style at follow up. Consideration should be given for the utility of CBT in relation to social phobia as people living with dystonia often experience scrutiny and negative responses such as stigma from others (116,117) so an approach that focusses on challenging 'incorrect beliefs', including performing behavioural experiments, may be counterproductive. An approach such as Acceptance and Commitment Therapy (ACT) may be more beneficial as the focus is on developing valued behaviours whilst accepting, rather than trying to change, any thoughts and feelings (118). ACT has shown promise as an intervention in long term health conditions and muscle disorders (119).

Given the role of body image and self-esteem in depression, interventions that focus on feelings of embarrassment and shame could be considered for this population. One such approach is to develop self-compassion, a gentle and kind way of relating to oneself during suffering as opposed to self-criticism (120), which can be developed using a Compassion Focussed Therapy approach (121). Self-compassion has been shown to decrease negative affect

in a study looking at participants with visible skin conditions (122). There is also evidence that it reduces body related distress and increases body appreciation in participants with breast cancer (123). Self-compassion has been associated with an increase in adaptive coping and decrease in maladaptive coping strategies in the context of chronic illness (124) suggesting that this may be a viable approach for these factors in those with dystonia.

### ***Limitations***

The BDI was the most commonly used measure of depression in the studies reviewed, however only one study engaged with the measure of the BDI in a way that considered the overlap in symptomology of dystonia (24). Scores on some of the measures may be artificially inflated due to their reliance on symptoms such as fatigue and sleep disruption which may be more related to the symptomology of dystonia rather than psychological distress (19). In addition, many of the tools used were screening rather than diagnostic which tend to focus on the two weeks prior to completion (125). This also makes it more difficult to draw comparisons between those studies that used screening measures and those which used the SCID which is a diagnostic tool. In addition, the wide variety of measures used outside of the BDI made comparisons and assimilation of the research difficult.

Another limitation was that the studies were all cross-sectional in design so causation cannot be inferred, and relationships may be bi-directional, as was shown in the relationship between body image and depression. The majority of the studies were also vulnerable to type 1 error due to the large number of statistical tests performed on the data, with only three accounting for this by either adjusting the significance level or utilising the Bonferroni correction. Therefore, caution should be applied when interpreting these results.

Furthermore, this review contained studies which examined a range of different types of dystonia. Whilst this is useful for an overall picture, the experiences of distress and psychological variables may differ depending on type and location of the condition.

### ***Future research***

This review highlighted that research on the psychological factors involved in anxiety and depression in those living with dystonia is limited. Research carried out with this population should remain critical of the measures used and take steps to ensure validity with this population. One option to consider might be the Depression Anxiety Stress Scale -21 (DASS-21) (126,127) which, although not used specifically with dystonia, has been used with other neurological conditions which present with a motor element (128,129). In addition, looking more closely at relationships between psychological factors and their effect on anxiety and depression utilising more complex statistical models and a longitudinal design will further advance understanding of the mechanisms involved in the development and maintenance of distress. An example of this would be a model that seeks to examine the relationship between BC, perceived disfigurement, self-depreciation and depression in those with cervical dystonia.

Social support is another area which warrants further research. It was only examined in one study within this review, however other reviews looking at psychological correlates of anxiety, depression and adjustment in both Parkinson's disease and multiple sclerosis identified social support and interactions as being related to levels of distress and adjustment (37,38). Social context and how it might impact on how one relates to oneself would also be a useful focus of future research, particularly given the increased amounts of stigma experienced by this population (116).

### ***Conclusion***

Drawing on a biopsychosocial understanding, this review aimed to identify research that had been conducted on psychological variables in relation to anxiety and depression and explore the strength of these relationships. A search of the literature revealed limited research had been done in this area, but included factors relating to distress such as coping styles, BC, self-esteem and personality traits, indicating a role for psychological variables and highlighting the need for more comprehensive research in this area. This also has clinical implications for the assessment and management of psychological distress in dystonia.

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**Table 1-1****Database search strategy**

Table 1-1 Database search strategy

CINAHL [97 hits]

Focus 1	Focus 2
Dystonia	Anxiety/ Depression
<ul style="list-style-type: none"> <li>• MH "Dystonia"</li> <li>• MH "Dystonic Disorders"</li> <li>• MH "Dystonia Musculorum Deformans"</li> <li>• MH "Focal Hand Dystonia"</li> <li>• MH "Meige Syndrome"</li> <li>• MH "Torticollis"</li> <li>• Free text "Dystonia musculorum deformans OR dystonic disorders OR generalised dystonia OR dystonia* OR torticollis* OR cervical dystonia OR spasmodic dysphonia* OR musician* cramp OR writer* cramp OR blepharospasm* OR meige* syndrome"</li> </ul>	<ul style="list-style-type: none"> <li>• MH "Depression"</li> <li>• MH "Affective Disorders"</li> <li>• MH "Anxiety"</li> <li>• Free text "anxiety or depression or psychiatric disorders or mental health or distress or stress or mood or wellbeing"</li> </ul>

MEDLINE [262 hits]

Focus 1	Focus 2
Dystonia	Anxiety/ Depression
<ul style="list-style-type: none"> <li>• MH "Dystonia"</li> <li>• MH "Dystonic Disorders"</li> <li>• MH "Dystonia Musculorum Deformans"</li> <li>• MH "Meige Syndrome"</li> <li>• Free text "Dystonia musculorum deformans OR dystonic disorders OR generalised dystonia OR dystonia* OR torticollis* OR cervical dystonia OR spasmodic dysphonia* OR musician* cramp OR writer*"</li> </ul>	<ul style="list-style-type: none"> <li>• MH "Depression"</li> <li>• MH "Anxiety Disorders"</li> <li>• MH "Anxiety"</li> <li>• MH "phobia, social"</li> <li>• Free text "anxiety OR depression OR psychiatric disorders OR mental health OR distress OR stress OR mood OR wellbeing"</li> </ul>

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cramp OR blepharospasm\* OR  
meige\* syndrome”

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PsycINFO [554 hits]

Focus 1	Focus 2
Dystonia	Anxiety/ Depression
<ul style="list-style-type: none"> <li>• DE "Torticollis"</li> <li>• DE "Tremor"</li> <li>• DE "Muscular Disorders"</li> <li>• DE "Muscle Spasms"</li> <li>• Free text “Dystonia musculorum deformans OR dystonic disorders OR generalised dystonia OR dystonia* OR torticollis* OR cervical dystonia OR spasmodic dysphonia* OR musician* cramp OR writer* cramp OR blepharospasm* OR meige* syndrome”</li> </ul>	<ul style="list-style-type: none"> <li>• DE "Anxiety Disorders"</li> <li>• DE "Death Anxiety"</li> <li>• DE "Generalized Anxiety Disorder"</li> <li>• DE "Obsessive Compulsive Disorder"</li> <li>• DE "Panic Attack"</li> <li>• DE "Panic Disorder"</li> <li>• DE "Phobias"</li> <li>• DE "Separation Anxiety Disorder"</li> <li>• DE "Trichotillomania"</li> <li>• DE "Anxiety"</li> <li>• DE "Major Depression"</li> <li>• DE "Anaclitic Depression"</li> <li>• DE "Dysthymic Disorder"</li> <li>• DE "Endogenous Depression"</li> <li>• DE "Late Life Depression"</li> <li>• DE "Reactive Depression"</li> <li>• DE "Recurrent Depression"</li> <li>• DE "Treatment Resistant Depression"</li> <li>• DE "Affective Disorders"</li> <li>• DE "Depression (Emotion)"</li> <li>• Free text “anxiety OR depression OR psychiatric disorders OR mental health OR distress OR stress OR mood OR wellbeing”</li> </ul>

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Web of Science [1876 hits]

Focus 1	Focus 2
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Dystonia	Anxiety/ Depression
<ul style="list-style-type: none"> <li>• Free text “Dystonia musculorum deformans OR dystonic disorders OR generalised dystonia OR dystonia* OR torticollis* OR cervical dystonia OR spasmodic dysphonia* OR musician* cramp OR writer* cramp OR blepharospasm* OR meige* syndrome”</li> </ul>	<ul style="list-style-type: none"> <li>• Free text “anxiety OR depression OR psychiatric disorders OR mental health OR distress OR stress OR mood OR wellbeing”</li> </ul>

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*Note:* MH = MeSH; DE = Descriptors. Searches were conducted on titles and abstracts. All terms for within focus were combined with OR. Focus 1 and 2 were combined with AND.

Table 1-2

## Characteristics of included studies

Table 1-2 Characteristics of included studies

Study/ country	Sample N (%m, Mean age); setting	Type of dystonia	Design & analysis	Psychological correlates	Depression/ Anxiety measures used	Findings
Cannito (1991), USA (48)	18 (0%, 48.53)	Spasmodic Dysphonia	Cross sectional, Between groups (control group = 18 healthy controls) and within group correlations	Somatic complaints	STAI; SDS	Somatic complaints were correlated with depression ( $r=.535$ ; $p<.05$ ) and state anxiety ( $r= .563$ ; $p<.05$ ), but not trait anxiety ( $r=.292$ ).
Gundel et al (2001), Germany (27)	116 (47%, 51.5); hospital	Spasmodic Torticollis	Cross sectional. Stepwise multiple regression	Depressive coping; body image dissatisfaction; incriminating life event	Structured clinical interview for DSM-IV; SPS (Social phobia)	Depressive coping ( $p<0.01$ ; odds ratio=10.8, 95% CI 3.0- 40.1) was the main predictor of current psychiatric comorbidity. Body image dissatisfaction was a predictor of social phobia ( $p=0.05$ ; odds ratio=2.4, 95% CI 1.0-6.3).
Hu et al (2013), USA (49)	145 (24.8%, 59.5); clinic; ns	Spasmodic Dysphonia	Cross sectional, correlations	Self-efficacy	HADS	High self-efficacy was negatively correlated with anxiety ( $r = -0.42$ ; $p<0.001$ ) and depression ( $r=0.42$ ; $p<0.001$ ) Disease specific self-efficacy was negatively correlated with anxiety ( $r = -$

Hu et al (2018), USA (50)	142 (25.4%, 59.2); clinic (from same sample as Hu at al. 2013)	Spasmodic Dysphonia	Cross sectional, logistic regression models	Self-efficacy & Disease Specific Self Efficacy	HADS	0.43; $p < 0.001$ ) and depression ( $r = -0.57$ ; $p < 0.001$ ). In a regression model A DSSE was a significant predictor of depression ( $p < 0.001$ ) and GSES was a significant predictor of anxiety ( $p < 0.01$ )
Ioannu et al (2014), Germany (46)	35 (80%, 45.5)	Focal Dystonia in Musicians	Cross sectional with exploratory cluster analysis	Perfectionism, Stress coping questionnaire	Competitive Trait Anxiety Inventory (CTAI)	All aspects of perfectionism and coping examined apart from 'flight tendency', correlated with 2 subscales of CTAI (self-doubt concern and somatic anxiety) ( $r = .7$ to $.5$ ; $p < .05$ )
Jahanshahi (1991), UK (43)	67 (52.3%, 53.3) ( a subgroup from the 1990 paper); mailing list	Spasmodic Torticollis	Cross sectional, correlation and stepwise multiple regression	Body concept, ways of coping, acceptance of illness, self-esteem, health locus of control	BDI	Maladaptive coping positively correlated with depression ( $r = 0.41$ ; $p < 0.001$ ) and adaptive coping was negatively correlated with depression ( $r = -0.27$ ; $p < 0.05$ ). Other correlations of depression significant to $p < 0.001$ were self-depreciation ( $r = 0.77$ ), self-worth ( $r = -0.71$ ), disfigurement ( $r = 0.5$ ), social support amount ( $r = -0.29$ ) and satisfaction ( $r = -0.36$ ), body concept ( $r = 0.72$ ) and belief in control of powerful

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<p>Jahanshahi &amp; Marsden (1990), UK (44)</p>	<p>85 (55.3%, 49.8); mailing list</p>	<p>Spasmodic Torticollis</p>	<p>Cross sectional, Between groups (control group = 49 with cervical spondylosis) and stepwise multiple regression</p>	<p>Body concept, personality traits</p>	<p>BDI</p>	<p>others (r = 0.54). A significance level of (p&lt;0.001) was chosen to account for the number of correlations. Stepwise multiple regression showed variance in depression explained by self-depreciation (59%), Satisfaction with social support (1.6%) and maladaptive coping (1.7%). Disfigurement rating (r = 0.39), extraversion (r = -0.42), neuroticism (r = 0.54), and body concept (r = 0.70) were correlated with depression (p&lt;0.05). In the regression model variance in depression scores was explained by extraversion (13.6%), disfigurement (13.5%) neuroticism (13%) and body concept (0.04%) when included with objective clinical measures. Neuroticism was significantly higher in those with anxiety (p&lt;0.001), depression (p&lt;0.01), panic disorder (p&lt;0.001) and social phobia (p&lt;0.001); openness</p>
<p>Lencer et al (2009), Germany (28)</p>	<p>86 (26.7%, 57.9f, 50m); clinic</p>	<p>Cervical Dystonia, Blepharospasm</p>	<p>Cross sectional, between groups (patients with primary focal dystonia affected by psychiatric disorder</p>	<p>Personality traits</p>	<p>Structured Clinical Interview for DSM-IV</p>	<p>Neuroticism was significantly higher in those with anxiety (p&lt;0.001), depression (p&lt;0.01), panic disorder (p&lt;0.001) and social phobia (p&lt;0.001); openness</p>

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			compared those who are not affected)			was significantly higher in those with panic disorder (p<0.05); Extraversion was significantly higher in those with social phobia (p<0.05). Self-esteem (r = -0.55; p<0.001), body concept (r = 0.16; p<0.001) and disfigurement (r = 0.15) were predictors of depression explaining 56%, 6% and 2% of the variance in depression respectively.
Lewis et al (2008), UK (24)	329 (31.3%, 56.1); epidemiological survey	Mixed dystonia subtypes	Cross sectional, correlation	Body concept, self esteem	BDI	
Mahajan et al (2018), USA (51)	208 (51.92%); dystonia coalition	Cervical Dystonia	Cross sectional, between groups analyses (those who met criteria for substance abuse compared with those who didn't)	Substance abuse	BDI-II, HADS, PHQ9, LSAS	Those who met the criteria for substance abuse scored higher on the PHQ-9 (p<0.05), BDI-II (P=0.005) and HADS (p<0.005) however there were no differences between the groups on the social anxiety scale (p=0.208).
Moraru et al (2002), Austria (52)	40 (50%, 44.1m, 48.1f); clinic	Cervical Dystonia	Cross sectional, between groups (comparison of depressed and non-depressed)	Scales on the Symptom Checklist (SCL-90)	BDI	Somatization, interpersonal sensitivity, obsessive compulsive and psychoticism scales of the SCL-90 were significantly different between patients with severe/moderate depression and non-depressed patients. (p<0.05)

Page et al (2007), UK (45)	276 (33.6%, 55); epidemiological survey- From same pool of participants as Lewis et al (2008)	Primary dystonia	Cross sectional, between groups and correlational	Subscales of SF36, Body concept, Self-esteem	BDI	BDI predicted body concept in stepwise multiple regression ( $r = 0.66$ ; $p < 0.001$ ) explaining 43% of the variance in body concept score.
Pekmezovic et al (2009), Serbia (53)	157 (40.76%, ST46.7, BS64, WC40.9); clinic	Spasmodic Torticollis, Blepharospasm, Writer's Cramp	Cross sectional, correlational	Psychosocial subscales of SF36	HDRS, HARS	Significant correlations between depression and social functioning across all groups ( $r = -0.621$ ; $p < 0.001$ ), and anxiety and social function for the torticollis ( $r = -0.525$ ; $p < 0.001$ ) and blepharospasm ( $r = -0.351$ ; $p < 0.05$ ) groups, but not writer's cramp ( $r = -0.386$ ; $p = 0.386$ ). Significant correlations between both depression and anxiety and role functioning (emotional) for the torticollis ( $r = -0.476$ ; $r = -0.489$ ; $p < 0.001$ ) and blepharospasm ( $r = -0.603$ ; $p < 0.001$ ; $r = -0.466$ ; $p < 0.05$ ) groups, but not writer's cramp ( $r = -0.433$ ; $p = 0.433$ ; $r = -0.446$ ; $p = 0.446$ ). All groups showed significant correlations between depression/anxiety and the

Perozzo et al (2016), Italy (54)	60 (50%); hospital	Blepharospasm and spasmodic torticollis (study also included 15 hemifacial spasm patients)	Cross sectional, Pearson's analyses	Sexual functioning	BDI, STAI	mental health subscale ( $r = -0.676$ ; $p < 0.01$ ; $r = -0.599$ ; $p < 0.01$ ). Depression was correlated with the total score on the SFI ( $r = 0.238$ , $p = 0.024$ ) and the infrequency subscale ( $r = 0.287$ , $p = 0.006$ ). The STAI-X1 correlated with the infrequency subscale ( $r = 0.306$ , $p = 0.003$ ) as did the STAI-X2 ( $r = 0.320$ , $p = 0.002$ ).
Scheidt et al (1996), Germany (47)	256 (40.7%, 49.1); hospital	Spasmodic Torticollis	Cross sectional, Correlation, stepwise multiple regression	Coping styles	SCL-90 (depression and anxiety subscales)	Depression and anxiety were both significantly correlated with depressive coping (depression $r = 0.68$ ; anxiety $r = 0.59$ ; $p < 0.001$ ), active problem focussed coping (depression $r = 0.13$ ; $p < 0.05$ ; anxiety $r = 0.16$ ; $p < 0.01$ ) and wishful thinking (depression $r = 0.34$ ; anxiety $r = 0.20$ ; $p < 0.001$ ).
Smit et al (2016), The Netherlands (55)	50 (54); clinic	Cervical dystonia	Cross sectional, Spearman's correlations	Psychosocial subscales of QoL measure	BDI, BAI	Depression was correlated with social functioning ( $r = -0.65$ ; $p < 0.01$ ), role limitation (emotional) ( $r = -0.61$ ; $p < 0.01$ ) and mental health ( $r = -0.71$ ; $p < 0.01$ ). Anxiety was not significantly correlated with social

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functioning ( $r = -0.25$ ;  $p < 0.2$ ) but was correlated with role limitation (emotional) ( $r = -0.45$ ;  $p < 0.01$ ) and mental health ( $r = -0.65$ ;  $p < 0.01$ ).

---

NOTE: BAI (Beck Anxiety Inventory); BDI (Beck Depression Inventory); CTAI (Competitive Trait Anxiety Inventory); HADS (Hospital Anxiety and Depression Scale); HARS (Hamilton Anxiety Rating Scale); HDRS (Hamilton Depression Rating Scale); LSAS (Liebowitz Social Anxiety Scale); PHQ-9 (Patient Health Questionnaire-9); (SCL-90 (Symptom Check List – 90); SDS (Self Rating of Depression Scale); SF-36 (Short Form Survey-36); SPS (Social Phobia Scale); STAI (State-Trait Anxiety Inventory).

**Table 1-3**  
**Quality appraisal**

Table 1-3 Quality appraisal

	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	Total
Cannito, 1991	1	1	0	1	0	0	0	1	1	1	1	1	0	0	1	1	1	0	1	0	12
Gundel et al 2001	1	1	0	1	1	1	1	1	1	1	0	0	1	1	0	0	1	1	1	1	15
Hu et al 2013	1	1	0	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	0	1	18
Hu et al 2018	1	1	0	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	0	1	18
Ioannou & Aktenmu llwe (2014)	1	1	0	1	1	1	0	1	1	1	1	1	0	0	1	0	1	1	1	1	15
Jahansha hi (1991)	1	1	0	1	1	1	1	1	1	1	1	1	1	1	1	1	1	0	1	0	17
Jahansha hi &	1	1	0	1	1	1	1	1	1	1	1	1	1	1	1	1	1	0	1	0	17

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Marsden (1990)																					
Lencer et al (2009)	1	1	0	1	1	1	1	1	1	1	1	1	0	0	1	1	1	1	1	0	16
Lewis et al (2008)	1	1	0	1	1	1	1	1	1	1	0	1	1	1	1	0	1	0	1	1	16
Mahajan et al (2018)	1	1	0	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	0	1	18
Moraru et al (2002)	1	1	0	1	1	1	0	1	1	1	1	1	0	0	1	0	1	0	0	0	12
Page et al (2007)	1	1	0	1	1	1	1	1	1	1	1	0	1	1	1	1	1	0	0	0	15
Pekmezovic et al (2009)	1	1	0	1	1	1	0	1	1	1	1	1	0	0	1	1	1	1	1	1	16
Perozzo et al (2016)	1	1	0	1	1	0	0	1	1	1	1	1	0	0	1	1	1	0	1	1	14
Scheidt et al (1996) <sup>1</sup>	1	1	0	1	1	1	1	1	1	1	1	1	0	0	1	1	1	0	0	0	14

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<sup>1</sup> This paper was a single on in a series describing a multicentre study so was examined alongside the first paper of the series which outlined the participants and methods..

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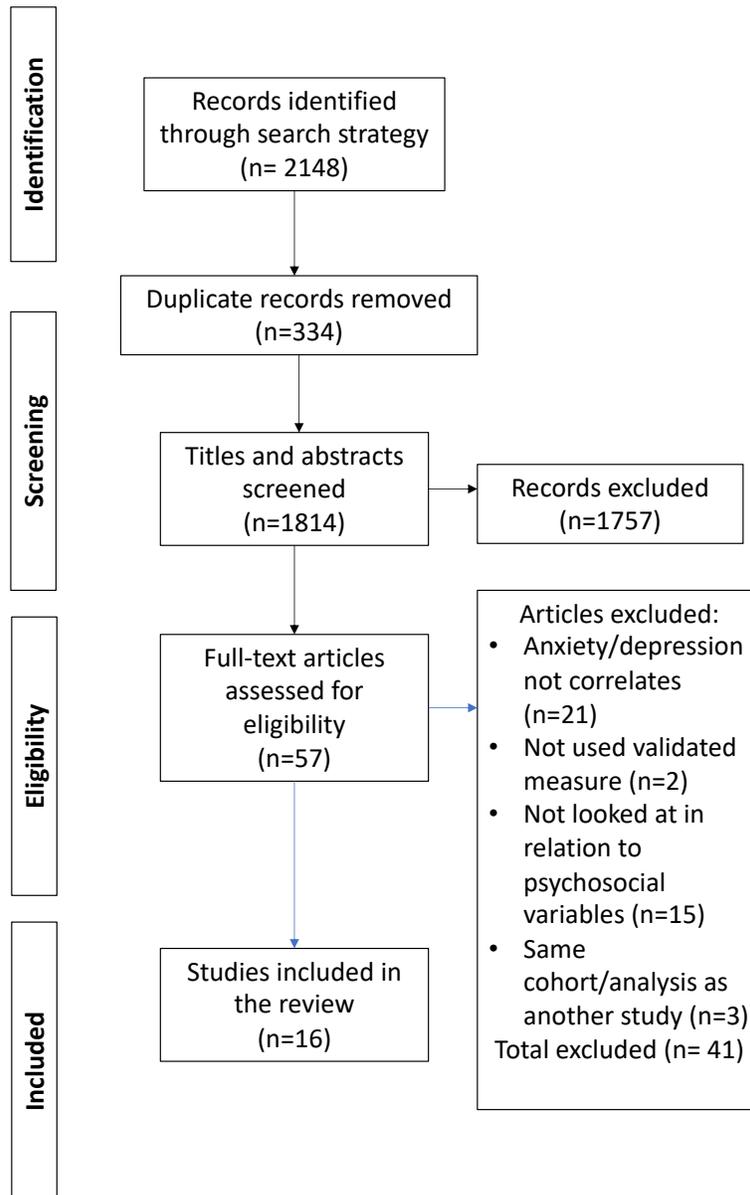
Smit et al (2016)	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	20
----------------------	---	---	---	---	---	---	---	---	---	---	---	---	---	---	---	---	---	---	---	---	----

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Note: The questions refer to the following sections of studies: 1= introduction; 2-11= method; 12-16 = results; 17-18 = discussion; 19-20 = other.

Full questions available in appendix 1.

**Figure 1-1: Study search flow chart**



**Appendix 1-1****Appraisal tool for Cross-Sectional Studies (AXIS tool)**

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**Questions**

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**Introduction**

1. Were the aims/objectives of the study clear?

**Methods**

2. Was the study design appropriate for the stated aim(s)?

3. Was the sample size justified?

4. Was the target/reference population clearly defined? (Is it clear who the research was about?)

5. Was the sample frame taken from an appropriate population base so that it closely represented the target/reference population under investigation?

6. Was the selection process likely to select subjects/participants that were representative of the target/reference population under investigation?

7. Were measures undertaken to address and categorise non-responders?

8. Were the risk factor and outcome variables measured appropriate to the aims of the study?

9. Were the risk factor and outcome variables measured correctly using instruments/ measurements that had been trialled, piloted or published previously?

10. Is it clear what was used to determine statistical significance and/or precision estimates? (e.g., p values, CIs)

11. Were the methods (including statistical methods) sufficiently described to enable them to be repeated?

**Results**

12. Were the basic data adequately described?

13. Does the response rate raise concerns about non-response bias?

14. If appropriate, was information about non-responders described?

15. Were the results internally consistent?

16. Were the results for the analyses described in the methods, presented?

**Discussion**

17. Were the authors' discussions and conclusions justified by the results?

18. Were the limitations of the study discussed?

**Other**

19. Were there any funding sources or conflicts of interest that may affect the authors' interpretation of the results?

20. Was ethical approval or consent of participants attained?

Total

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## Appendix 1-2

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*Updated 3-06-2020*

**Section Two: Research Paper**

**Stigma, coping strategies and wellbeing in individuals with cervical dystonia**

Word count (excluding references, tables and appendices): 7900

Helen Gowling

Doctorate in Clinical Psychology

Division of Health Research, Lancaster University

Prepared for submission to *Psychology & Health* (see appendix 2-1)

**All correspondence should be sent to:**

Helen Gowling  
Doctorate in Clinical Psychology  
Lancaster University  
Furness College  
United Kingdom  
LA1 4GY

Email : [h.gowling@lancaster.ac.uk](mailto:h.gowling@lancaster.ac.uk)  
Tel: 01524 59297

## Abstract

**Objective:** The purpose of this study was to investigate whether coping strategies (adaptive vs maladaptive) mediate the relationship between stigma and wellbeing in people living with cervical dystonia.

**Design:** A cross-sectional design was used. Adults with cervical dystonia were invited to take part in a survey. 114 individuals completed quantitative measures of stigma and coping strategies and a holistic exploration of the concept of wellbeing (including health-related quality of life, depression, anxiety, stress, and a wellbeing scale). Data were analysed using mediation regression analyses.

**Main findings:** Mediation regression analyses indicated that maladaptive coping strategies (e.g. substance use, behavioural disengagement) mediated the relationship between stigma and wellbeing. Adaptive coping strategies (e.g. acceptance, humour) did not mediate the relationship between stigma and wellbeing.

**Conclusion:** These findings suggest that maladaptive coping may play an important role in explaining the relationship between stigma and some aspects of wellbeing in cervical dystonia. Coping strategies should be considered within clinical settings to target the relationship between stigma and wellbeing. Interventions which focus on different aspects of maladaptive coping (e.g. acceptance and commitment therapy for people who are using strategies such as denial or experiential avoidance) may be helpful to improve wellbeing.

*Keywords:* Cervical dystonia, spasmodic torticollis, stigma, coping, psychological wellbeing

## Introduction

Dystonia is a movement disorder which involves involuntary, sustained muscle contractions leading to repetitive movements and abnormal postures (1). Studies have shown that it affects approximately 0.01-0.03% of the population (2,3) and is the third most prevalent movement disorder after Parkinson's disease and essential tremor (4). Although the neurobiology of this condition is still not fully understood (5), it has been proposed that dystonia should be classified on two axes (6). Axis I includes clinical characteristics such as age of onset and body distribution, which is broken down into the following: focal (affecting one body region), segmental (affecting two or more adjoining body regions), multifocal (affecting two non-adjoining or more body regions), generalised (affecting the trunk and two other body regions), and hemidystonia (body regions affected are more focussed on one side). Axis II includes what is known of the aetiology, including whether it is inherited, acquired or idiopathic dystonia (6). Cervical dystonia (CD), also known as spasmodic torticollis, is the most common focal dystonia and is characterised by uncontrolled twisting, turning and tilting of the head caused by involuntary and often painful contractions of the neck muscles (7).

In addition to the physical implications, people with dystonia report a range of psychological difficulties that impact on their day to day lives. Reports in the literature estimate that lifetime prevalence of anxiety and depression for people living with dystonia varies between 12%-71% (8). There is debate over whether psychological distress is a result of the underlying neurobiology associated with the condition, or whether it is secondary to the impact of living with a chronic health condition (9,10). There is evidence to suggest it could be a combination of both, and that a biopsychosocial framework should be adopted when trying to understand psychological distress in this population (11), particularly as distress can impact overall health

related quality of life (HRQOL) (12). HRQOL broadly looks at how a condition impacts on an individual across physical, social and psychological domains (13,14).

A range of factors have been shown to correlate with distress and HRQOL, including body image (15–19), social participation (7,16), self-esteem (7,16,17) and coping strategies (15,16,20,21). In studies of other movement disorders, such as Parkinson's disease, stigma has also been found to play a key role in HRQOL, distress and wellbeing (22,23).

Stigma is a vital aspect of health research and was originally defined as a feeling of being shunned by others for possessing a distinguishing characteristic that is less valued than the social norm (24). Stigma can be broken down into two main areas: enacted stigma, which is related to discrimination and attitudes from the public; and perceived stigma, which is an internalising of enacted stigma by an individual, often leading to feelings of shame and can often occur without many instances of enacted stigma (25). Theoretical frameworks of stigma were further expanded by Link & Phelan (26) who recognised the role of power inequalities and the relational context that enables stigma to occur.

Perceived stigma has been shown to lead to poorer quality of life (QOL), negatively impact health-related outcomes and increase social isolation in people with mental health difficulties (27–29). Studies of enacted stigma involving people with epilepsy show that frequency of enacted stigma is associated with increased psychological distress and a poorer QOL (30–32). Stigma has also been found to predict psychological outcomes in people with neurological conditions who present with movement disorders such as Parkinson's disease and multiple sclerosis (23,33,34). Visible characteristics, such as tremors or muscle contractions in dystonia, can result in stigmatising social experiences alongside the physical and emotional

impact of the condition (35). This in turn can exacerbate the psychological, physical and social aspects of the condition (36) and may contribute towards a decrease in overall QOL (37).

A 2001 study examining the role of stigma in cervical dystonia found that the majority of participants reported experiencing 'some' or 'severe' stigma relating to their condition (38). The questionnaire used to examine stigma was created following interviews with participants.

Although this meant that it had good face validity and was focussed on what was important to the 10 participants initially interviewed, it is unclear how generalisable this could be to other people living with dystonia. An additional study looking at the perceptions of people without dystonia used a scale developed in line with that used by Papathanasiou et al. (38) and revealed stigmatising attitudes towards those with cranial and cervical dystonia (39). This suggests that enacted stigma may be a part of daily life for this population, particularly those with cervical dystonia. Stigma was also a strong theme in recent qualitative research on the experience of living with dystonia, with the findings suggesting that stigma impacted on the navigating of health services, social connectedness and views of the self (40).

The role of stigma in relation to psychological outcomes for people with dystonia is poorly understood. One study found a significant relationship between stigma and both the mental and physical aspects of QOL in people with cervical dystonia (7). Another study found no significant relationship between stigma and HRQOL for people with segmental dystonia (41) but had a small sample size (n=28). Furthermore, both studies used a stigma scale designed for people with bowel cancer (42) which had not been validated for dystonia. Despite some emerging evidence for the role of stigma in psychological well-being in dystonia, the picture is still unclear and may indicate that there are other factors that could be influencing the relationship between the two concepts.

Research focussing on other groups who experience stigma has considered the ‘minority stress paradigm’ (43), which conceptualises stigma as a stressor above and beyond what non-stigmatised people endure, leading to difficulties with mental health. This paradigm was further developed into a ‘psychological mediation framework’ (44), which integrated the theory of minority stress with psychological processes that may result from being stigmatised, and therefore contribute to psychological distress. One of these proposed processes was coping strategies.

Coping can be defined as the behavioural, emotional and cognitive strategies employed to manage certain stressors (45). It is suggested that coping has an important role to play in the adaptation needed due to changes in physical health (46). Similarities have been noted across different disease processes in how people cope with the stress of changes in ability, and the impact that this then has on emotional distress (47). There are over 400 coping strategies which have been classified in various ways within the literature, such as ‘emotion focussed’, ‘problem focussed’, ‘approach’, ‘avoidance’, ‘adaptive’ and ‘maladaptive’, without a shared consensus on the most appropriate classification (48). In line with research drawn on for the current study, the terms ‘maladaptive’ and ‘adaptive’ will be used. It is important to note however, that using these terms is not meant to encourage simplistic thinking about ‘bad’ or ‘good’. It is recognised that maladaptive coping is often a functional adaptation to chronic stress (49) and that a two factor model may not fully capture the complexities related to coping strategies. However, within the literature, the use of a two factor model has provided insight into the impact of coping strategies on levels of distress (50,51) suggesting there is utility in this approach. In particular, maladaptive coping strategies have a greater relationship with distress, whereas adaptive strategies have a stronger relationship with wellbeing (50).

There is emerging evidence that the experience of stigma can lead to coping behaviours which could be considered maladaptive, such as disengagement and substance use, which in turn can impact on psychological wellbeing and QOL (52). Coping strategies have also been shown to be related to distress in dystonia, with strategies considered maladaptive being related to higher levels of depression and anxiety (15,16,20,21).

Wellbeing has not been examined in dystonia, apart from as an outcome in a singular treatment study (53). Wellbeing is usually conceptualised as a combination of functioning well and positive affect (54) although it is generally poorly defined within the literature (55). However, wellbeing has been shown to be an important predictor of physical health in longitudinal research (56,57). It is also suggested that if we only examine distress then there is a risk that this might leave important gaps in our understanding of health and QOL (58).

Although there is a suggested relationship between stigma and QOL, and between coping strategies and psychological distress, to date the relationship between stigma, coping and psychological outcomes has not been examined in people with dystonia. Consequently, this study will investigate, via survey, the relationship between stigma and psychological wellbeing, then also investigate whether coping mediates this relationship in a cohort of people with cervical dystonia. The focus will be on a community sample of people with cervical dystonia as this is the most common type of dystonia. It is hoped that by furthering the understanding of these concepts, clinicians will be able to better support the wellbeing of those living with this chronic condition. This in turn could have an impact on overall QOL.

For the purposes of this study, well-being is characterised by low levels of distress labelled as anxiety, stress and depression, as well as an elevated score on a measure of well-being. A measure of HRQOL specific to cervical dystonia is also included to gain a holistic

picture of satisfaction across a variety of life domains. The model being tested will be of theoretical interest and has potential implications for the field of clinical psychology both on an individual level within formulations and at a more systemic and societal level (59).

Based on previous research the study hypotheses were as follows:

1. Higher levels of stigma will be associated with higher maladaptive but lower adaptive coping strategies.
2. Higher levels of stigma will be associated with lower levels of psychological well-being, increased levels of distress (depression, anxiety and stress) and reduced HRQOL.
3. The use of maladaptive coping strategies will be associated with increased distress, lower wellbeing and lower HRQOL.
4. Maladaptive coping will mediate the relationship between stigma and psychological well-being, levels of distress and HRQOL.
5. Adaptive coping will mediate the relationship between stigma and psychological well-being.

## **Method**

### ***Design***

This study was a cross-sectional survey using quantitative measures. The data were quantitatively examined, and non-parametric bivariate correlations were carried out. Following this, a parallel mediation analysis was conducted using Hayes process tool (60) within SPSS to test the theoretical model that coping strategies mediate the relationship between stigma and psychological wellbeing (including distress) and HRQOL (see figure 2-1).

[Figure 2-1 about here].

### ***Participants***

Participants were recruited via two dystonia charities: The Dystonia UK (<https://www.dystonia.org.uk/>) and Dystonia Ireland (<http://www.dystonia.ie/>). Recruitment took place between 3<sup>rd</sup> September 2019 and 27<sup>th</sup> January 2020 via advertisements on the charities' social media profiles. Two participants contacted the researcher to request paper copies of the survey which were then returned in pre-paid envelopes and manually inputted into the survey software Qualtrics. The rest of the participants completed the survey online. A total of 143 people completed the survey. The data from 29 participants were removed due to missing data (see analysis for details). This left 114 participants to be included in the study.

Calculations were conducted prior to recruitment to determine sample size required. For a medium effect (0.36) in both paths (stigma to coping ( $\alpha$ ) and coping to psychological outcomes ( $\beta$ )) using a bias-corrected bootstrap mediation model, approximately 71 participants were required for 80% power, rising to 115-116 if the  $\alpha$  or  $\beta$  path coefficient has a small-medium effect (0.26) (61). Previous research looking at the relationship between stigma and coping have found small-medium effects for adaptive coping and large effects for maladaptive coping, suggesting powering the study for a small-medium effect should be sufficient (51). It is not possible to take the effect size for  $\beta$  from previous research as this requires the identical model (and the author is not aware of such research). Therefore, the sample size of 114 was deemed acceptable.

### ***Inclusion criteria***

Individuals were eligible to take part if they:

- Self-reported a diagnosis of cervical dystonia.

- Were aged 18 or over.
- Could complete the measures alone or with support.
- Had sufficient understanding of written English to take part (as the questionnaires were in English).

### ***Exclusion criteria***

Potential participants were excluded if they:

- Had dystonia following a serious injury.
- Had another significant illness/condition that affected their visible appearance.

### ***Procedure***

The study was designed in consultation with two members of Dystonia UK who provided feedback via email on the content and accessibility of the survey. Adaptations were made based on their feedback, including the change of wording on some of the questions and explanations. Once ethical approval had been obtained the study was advertised on the Facebook and Twitter pages for Dystonia UK and Dystonia Ireland as well as in communications that were cascaded to local groups. People who followed the link were presented with an information and consent page explaining the purpose of the study, and invited to complete an online survey (using Qualtrics, a web-based survey tool) or to contact the researcher to request a paper copy to be sent via post (for information page and survey see section 4: Ethics and Appendices). Data from the online survey were gathered electronically, and data from the hard copies were inputted immediately into the electronic dataset. The hard copies of the questionnaire were safely and immediately destroyed.

### ***Materials***

This study utilised an online survey which was also available in paper form. Alongside the validated measures it also asked questions regarding demographic and clinical information to describe the sample. The demographic questionnaire asked about age, gender, ethnicity, work status, relationship status and living arrangements. The clinical questionnaire asked about age of onset, duration, time since diagnosis, medication, disease severity, whether the individual was receiving Botulinum toxin treatment, any other treatments and any other health conditions.

### ***Validated measures***

#### ***Predictor variable***

##### ***Stigma***

The predictor variable, stigma, was measured by the 24-item Stigma Scale for Chronic Illness (SSCI). This scale was developed for people with chronic neurological disorders and measures both felt stigma and enacted stigma (62). This scale is divided into two subscales of internalised stigma and enacted stigma. All questions are answered using a Likert scale from one (never) to five (always). The total score of both scales measures overall stigma and can range from 24-120, with a higher score suggesting more stigma. It has been shown to have a good level of internal consistency (Cronbach's alpha = 0.96) (62) and good content validity (63).

#### ***Mediator variables***

##### ***Coping***

The two mediator variables in this study were adaptive coping and maladaptive coping. These were both measured using the brief COPE (64). This has 14 subscales each including two items. Although the subscales of the brief COPE have been categorised in various ways in the literature, this study used the two-category model developed by Meyer (50), which divides the subscales into adaptive and maladaptive coping strategies. All questions were answered using a

Likert scale from one (I haven't been doing this at all) to four (I've been doing this a lot).

Adaptive coping included the subscales 'active coping', 'use of emotional support', 'use of instrumental support', 'positive reframing', 'planning', 'humour', 'acceptance' and 'religion'.

This scale gave scores ranging from 16-64 with a higher score indicating more frequent use of these strategies. Maladaptive coping included the subscales 'self-distraction', 'denial',

'substance-use', 'behavioural disengagement', 'venting' and 'self-blame'. This scale gave scores ranging from 12-48 with a higher score indicating more frequent use of these strategies.

Although not used specifically in dystonia, the brief COPE has been used with other neurological conditions such as Parkinson's disease (65) and multiple sclerosis (66). For the current study it showed good reliability with Cronbach's alpha values of 0.81 for the maladaptive subscale and 0.84 for the adaptive subscale

### ***Outcome variables***

#### ***Well-being, distress and HRQOL***

Three separate measures were included to address the outcome of wellbeing. The first was the short version of the Depression Anxiety Stress Scale-21 (DASS-21) (67,68). This is a 21-item scale with three subscales looking at stress, anxiety and depression. Each subscale has 7 questions which are rated on a Likert scale from zero (not at all) to three (most of the time). Each subscale score ranges from 0-21 with a high score indicating higher levels of stress, anxiety or depression. Although not used previously with people living with dystonia, it has been used with other neurological conditions which present with movement disorders such as Parkinson's disease (69) and multiple sclerosis (70). It has good reliability with subscale Cronbach's alpha values of 0.81 (depression), 0.89 (anxiety) and 0.78 (stress) (71).

The Warwick-Edinburgh Mental Well-being Scale (WEMWBS) (72) is a 14-item validated measure of wellbeing which focusses on positive aspects of mental health. This has been used as an outcome in a previous study looking at the effects of a cognitive behavioural intervention for people living with dystonia (53) and more recently in a large survey of people living with Parkinson's disease (73). In addition, it is considered easy to complete and useful for general population samples. Each item is scored using a Likert scale from one (never) to five (always) giving a total score range of 14-70. In this scale a higher score indicates higher levels of wellbeing. The WEMWBS has been shown to have a strong internal consistency and a high Cronbach's alpha of 0.89 (72).

The measure of overall HRQOL was the Cervical Dystonia Impact Profile (CDIP-58) (74) which is a 58-item scale measuring the impact of CD across eight domains: head and neck symptoms, pain and discomfort, upper limb activities, annoyance, sleep, walking, mood and psychosocial functioning. A higher score shows a lower HRQOL. This measure has been shown to have good validity and reliability with a Cronbach's alpha of 0.92 (75).

To help describe the sample, disease severity was also explored using the Functional Disability Questionnaire (76). This is a 27-item scale developed to measure the impact of dystonia on activities of daily living with a higher score indicating higher levels of disability. It has been suggested by the original authors of the measure that scores above the median of 42 indicate a moderate to severe level of disability relating to dystonia (16). The construct and concurrent validity of the scale, along with test-retest reliability and internal consistency have been shown to be high with a Cronbach's alpha of 0.92 (76).

### *Ethical Considerations*

Ethical approval was obtained for this study via the Lancaster University Faculty of Health and Medicine Research Ethics Committee (reference FHMREC18109 – see section 4: Ethics and appendices for ethical approval letter). An amendment was applied for (and granted) in October 2019 to expand the recruitment strategy to enable both charities to be able to advertise through all channels available to them and to be able to approach other dystonia relevant online organisations to request the advert be shared through their channels. Dystonia UK also requested a video that they could use to disseminate information about the study.

Efforts were made to consider any potential distress that may be caused by participating in this study. Contact details were given for places that could provide support should any issues arise. Additionally, it was made clear prior to starting the study and providing informed consent that individuals could stop at any time during the survey. Although, due to the anonymity of participation, their data could not be removed after they had agreed to take part. This information was provided before informed consent was obtained to be included in the study.

### *Data analysis*

One hundred and forty-three participants opened the survey and completed the consent process. Out of these, 13 had only completed the demographics and a further four had stopped after only completing the first measure. Additionally, five participants had not completed any of the measures of wellbeing. These data were therefore removed from the dataset (n=22) leaving 121 participants. A missing values analysis was conducted, and one case was missing multiple (more than two) data points from multiple measures, so this case was also removed from analysis. Six further participants had missed completing full scales that were essential to the analysis (SSCI; n=3, DASS21; n=3) so these were also removed leaving 114 participants. The

missing SSCI data may have been due to an error with the survey software, and the DASS21 was situated towards the end of the survey so people may have exited before completing it. There were no significant differences in the demographic and clinical characteristics between those who completed all the measures and those who did not. Of the remaining cases, none were missing more than two data points from any measure, so they were retained, and mean substitution was used to impute the missing data points.

Absolute values for z-scores were calculated for each variable to check whether they were consistent with a normal distribution (77). This analysis identified probable outliers (with scores of  $>2.58$ ) in three of the variables: two cases on the SSCI (1.7%), two cases on the maladaptive subscale of the brief COPE (1.7%) and one case on the anxiety subscale of the DASS21 (0.8%). ‘Winsorising’ the data on these scales had little effect on the results of the analyses so the original data were retained.

Data distributions were inspected visually for normality using histograms. The ‘maladaptive coping’, ‘depression’, ‘anxiety’ and ‘stress’ data were all skewed towards lower scores, so Spearman’s rho correlation coefficients were calculated to assess the direction and strength of relationships between predictor, mediator and outcome variables. Before conducting the mediation analyses, the linear and multiple regression scatterplots corresponding to each analysis in the mediation were also visually inspected to test the assumptions of linearity and homoscedasticity of residuals, with standardised residuals plotted against standardised predicted values (77). Q-Q plots were used to assess the assumption of normality of error distributions (77). All relationships appeared to respect the assumptions of linearity, homoscedasticity of residuals and normality of error distributions.

Finally, a series of mediation analyses were conducted using Hayes’ Process Tool (78). In

each analysis, 5000 bootstrap samples were used to estimate the confidence intervals, enabling it to cope with non-normal data distributions (61). In all models, stigma as measured by the SSCI, was the independent variable (IV) and two types of coping strategies (adaptive and maladaptive as measured by the Brief COPE) were tested together as parallel mediators. In model 1, HRQOL as measured by the CDIP-58 was used as the dependent variable (DV). In model 2, wellbeing as measured by the WEMWBS was used as the DV. In model 3 stress as measured by the stress subscale of the DASS-21 was used as the DV. In model 4, anxiety as measured by the anxiety subscale of the DASS-21 was used as the DV. In model 5 depression as measured by the depression subscale of the DASS-21 was used as the DV. The indirect effect of coping style was deemed significant if the 95% bootstrap confidence interval did not contain zero (60).

## RESULTS

A summary of participant characteristics is included in Table 2-1.

[Table 2-1 about here]

Of the 114 participants in the sample, 21 identified their gender as being male (18%) and 93 identified as female (82%). The mean age of the sample was 52.39 years and ages ranged from 24-77 years. Most participants (111; 97%) identified their ethnicity as white (English, Irish, British, Scottish, Welsh, Other), one as mixed white & black Caribbean and two as 'other'. Half the participants (57) reported being in either full time or part time work, 19 (17%) were retired and 38 (33%) reported they were not currently working.

### *Clinical characteristics*

The participants reported a mean age of symptom onset of 34.49 years (*SD* 12.86) and a mean age of diagnosis of 42.60 years (*SD* 11.89). Most of the participants (88%) reported they

had received botulinum toxin injections to manage the symptoms of cervical dystonia and 63 participants (55%) reported taking prescribed medication to manage symptoms. Approximately half of the participants (54%) reported having comorbid physical or mental health conditions. For those that completed the FDQ (n=111) the mean score was 72.27 (*SD* 20.9) and a range of 25-12.

### ***Measures of stigma, coping and wellbeing***

Table 2-2 shows the means, standard deviations (*SD*) and Cronbach's alpha of psychometric measures for the sample.

[Table 2-2 about here]

[Table 2-3 about here]

Table 2-3 shows the frequencies and cut offs for measures of distress using the DASS-21. These results suggest that 48.3% (n=55) of the sample reported moderate to extremely severe levels of depression, 55.3% (n=63) showed moderate to extremely severe levels of anxiety, and 36.8% (n=42) of the sample reported moderate to extremely severe levels of stress. Additionally the results from the WEMWBS, which indicate levels of wellbeing, showed that 43% (n=49) scored lower than 40 which is the level that UK National Health Service direct uses as a cut off score to indicate lower levels of wellbeing (79).

The mean score for adaptive coping was 36.1 and for maladaptive coping was 22.6. Although there are no cut offs for this scale, higher scores indicate more frequent use of the different coping strategies.

The mean score for the measure of stigma for this sample was 61.33. Higher scores indicate increased levels of reported stigma. These scores were higher than the normative sample mean of 42.7 which was found in participants with a range of other neurological conditions (62).

The CDIP-58 scaled mean was 50.24 which is similar to that seen in other research on cervical dystonia (80,81) and suggests a significant impact of CD on overall HRQOL.

### ***Correlational analyses***

Non-parametric bivariate correlations indicated that the relationships between all the scales, apart from the adaptive subscale of the Brief COPE, were significant at the  $p < 0.01$  level with medium to large effect sizes (see table 2-4). Specifically, higher levels of stigma correlated with more frequent maladaptive coping ( $r = .637$ ), lower HRQOL ( $r = .641$ ), lower levels of wellbeing ( $r = -.488$ ), higher levels of stress ( $r = .529$ ), higher levels of anxiety ( $r = .484$ ) and higher levels of depression ( $r = .589$ ), but not use of adaptive coping ( $r = .04$ ;  $p > 0.05$ ). More frequent use of maladaptive coping strategies correlated with lower HRQOL ( $r = .528$ ), lower levels of wellbeing ( $r = -.427$ ), higher levels of stress ( $r = .559$ ), higher levels of anxiety ( $r = .538$ ), and higher levels of depression ( $r = .631$ ). More frequent use of adaptive coping strategies only correlated with an increase in wellbeing with a medium effect size ( $r = .331$ ,  $p < .01$ ).

Gender was found to significantly correlate with the anxiety subscale of the DASS-21 ( $r = .198$ ;  $p < 0.05$ ) and the CDIP-58 ( $r = .219$ ;  $p < 0.05$ ) with females scoring higher on both of these measures with a small effect size. This suggests that females in this sample had higher levels of anxiety and a lower HRQOL.

[Table 2-4 about here]

### ***Mediation regression analyses***

Five parallel mediation models (60,82) were used to examine the indirect effect of both adaptive and maladaptive coping on different measures of wellbeing: HRQOL, CDIP-58 (Model 1); wellbeing, WEMWBS (Model 2); stress, DASS21-S (Model 3); anxiety, DASS21-A (Model

4); and depression, DASS21-D (Model 5) (see Figure 2-2). For a summary of results see Table 2-5.

[Figure 2-2 about here]

[Table 2-5 about here]

### ***Model 1 – HRQOL***

Results from the parallel mediation analysis indicate that stigma was indirectly related to HRQOL through its relationship with maladaptive coping. Increased stigma was related to more frequent use of maladaptive coping strategies ( $a_2=.246, p<.001$ ), and more frequent maladaptive coping was subsequently related to a lower HRQOL ( $b_2=2.006, p <.01$ ). Mediation analysis indicated that the 95% confidence interval of the indirect effect through maladaptive coping ( $a_2b_2=.492$ ) was entirely above zero (0.136 to 0.857). In contrast, increased stigma was not related to more frequent use of adaptive coping strategies ( $a_1=.003$ ) and adaptive coping was not related to HRQOL ( $b_1=-.456$ ). The indirect effect through adaptive coping was not significantly different to zero ( $a_1b_1=-.001$ ; 95%CI -.060 to .074; see table 2-E for the effect sizes associated with these pathways). The direct effect between stigma and HRQOL was found to remain significant when controlling for the effect of the mediational variables of coping strategies ( $c' = 1.367, p<.001$ ).

### ***Model 2 – wellbeing***

The analysis indicated that stigma was indirectly related to wellbeing through its relationship with maladaptive coping. Increased stigma was related to more frequent use of maladaptive coping strategies ( $a_2 = .246, p<.001$ ), and more frequent maladaptive coping was subsequently related to a lower wellbeing ( $b_2 = -.549, p<.005$ ). Mediation analysis indicated that the 95% confidence interval of the indirect effect through maladaptive coping ( $a_2b_2 = -.135$ ) was

entirely below zero (-.231, to -.050). In contrast, increased stigma was not related to more frequent use of adaptive coping strategies ( $a_1=.003$ ) but more frequent adaptive coping was related to higher wellbeing ( $b_1= .372, p<.001$ ). However, the indirect effect through adaptive coping was not different to zero ( $a_1b_1= .001$ ; 95%CI -.037 to .038). The direct effect between stigma and wellbeing remained significant when controlling for the effect of the mediational variables of coping strategies ( $c' = -.194, p<.005$ ).

### ***Model 3 – stress***

The analysis indicated that stigma was indirectly related to stress through its relationship with maladaptive coping. Increased stigma was related to more frequent use of maladaptive coping strategies ( $a_2 = .246, p<.001$ ), and more frequent maladaptive coping was subsequently related to higher levels of stress ( $b_2 = 0.371, p<.001$ ). Mediation analysis indicated that the 95% confidence interval of the indirect effect through maladaptive coping ( $a_2b_2 = .091$ ) was entirely above zero (.050 to .137). In contrast, increased stigma was not related to more frequent use of adaptive coping strategies ( $a_1=.003$ ) and adaptive coping was not related to stress ( $b_1= -.020$ ). The indirect effect through adaptive coping was not different to zero ( $a_1b_1= .000$ ; 95%CI -.004, .005). The direct effect between stigma and stress remained significant when controlling for the effect of the mediational variables of coping strategies ( $c' = .061, p<.05$ ).

### ***Model 4 – anxiety***

The analysis indicated that stigma was indirectly related to anxiety through its relationship with maladaptive coping. Increased stigma was related to more frequent use of maladaptive coping strategies ( $a_2 = .246, p<.001$ ), and more frequent maladaptive coping was subsequently related to higher levels of anxiety ( $b_2 = 0.308 p<.001$ ). Mediation analysis indicated that the 95% confidence interval of the indirect effect through maladaptive coping ( $a_2b_2$

= .076) was entirely above zero (.035 to .116). In contrast, increased stigma was not related to more frequent use of adaptive coping strategies ( $a_1 = .003$ ) and adaptive coping was not related to anxiety ( $b_1 = -.030$ ). The indirect effect through adaptive coping was not different to zero ( $a_1 b_1 = .000$ ; 95%CI -.005, .005). The direct effect between stigma and anxiety remained significant when controlling for the effect of the mediational variables of coping strategies ( $c' = .091, p < .005$ ).

#### ***Model 5 – depression***

The analysis indicated that stigma was indirectly related to depression through its relationship with maladaptive coping. Increased stigma was related to more frequent use of maladaptive coping strategies ( $a_2 = .246, p < .001$ ), and more frequent maladaptive coping was subsequently related to higher levels of depression ( $b_2 = .489, p < .001$ ). Mediation analysis indicated that the 95% confidence interval of the indirect effect through maladaptive coping ( $a_2 b_2 = .120$ ) was entirely above zero (.075 to .172). In contrast, increased stigma was not related to more frequent use of adaptive coping strategies ( $a_1 = .003$ ) but more frequent adaptive coping was related to lower levels of depression ( $b_1 = -.135, p < .005$ ). The indirect effect through adaptive coping was not different to zero ( $a_1 b_1 = .000$ ; 95%CI -.016, .014). The direct effect between stigma and depression remained significant when controlling for the effect of the mediational variables of coping strategies ( $c' = .091, p < .01$ ).

#### **Discussion**

The current study aimed to examine associations between stigma, coping and psychological wellbeing for people living with CD. The findings suggested that higher levels of stigma were related to more frequent use of maladaptive coping strategies, lower wellbeing, lower HRQOL and higher levels of distress. More frequent use of maladaptive coping strategies

were related to lower levels of wellbeing, lower HRQOL and higher levels of distress. Adaptive coping was not related to stigma or distress, but more frequent use of adaptive coping strategies were related to higher levels of wellbeing. In all the mediation models, maladaptive coping mediated the relationship between stigma, HRQOL and wellbeing. Adaptive coping did not mediate the relationship between stigma and any of the outcomes.

This study was conducted with a cohort of adults living with CD. The results suggested that, according to the cut-offs on the DASS, 48.3% of the current sample had moderate to extremely severe depression, 55.3% had moderate to extremely severe anxiety and 36.8% had moderate to extremely severe levels of stress. These rates of distress are towards the high end of what other studies specifically looking at CD have reported, with rates of depression ranging from 16.4%-52.3% (15,16,76,83,84) and anxiety ranging from 21.7%-50% (15,83,84).

The mean wellbeing scores for this sample (41.68) were lower than the general population (49.7-52.3 across England and Scotland) (85), lower than a large scale survey of people with Parkinson's disease (45.2) (73) and a baseline measure in a dystonia cognitive behavioural therapy (CBT) study (44.7) (53). This suggests that the current sample might experience lower wellbeing than would typically be seen in dystonia. However, as wellbeing has not been thoroughly examined in this population, and the study did not have a control group to permit direct comparisons, more research is needed to further understand how dystonia impacts this. In addition, the scores on the FDQ suggested that the CD population in the current study self-reported high levels of disability with a mean score of 72.27. This is higher than has been indicated in some other research using the same scale in dystonia (16,17) which reported means of 44.1 and 50.3 respectively, but lower than one more recent study (mean 82.6) (86). The high

levels of disability in the current study may be one of the factors relating to the levels of distress and wellbeing reported in the current study.

The mean scores for both adaptive and maladaptive coping were lower than found in a student population using the same measure and categorisation (51), suggesting that those who took part in this study used both adaptive and maladaptive strategies with less frequency than the student population. The mean score for the measure of stigma for this sample was 61.33. This is higher than has been found in other populations such as stroke (mean = 45.21) and multiple sclerosis (mean range 36.45–47.44) (87,88), suggesting that those in the current study experience a high amount of stigma. It is also the first time this scale has been used specifically with CD. However, it is important to consider the findings relating to distress and stigma with caution due to the data being collected via an online questionnaire and the lack of a comparison group, so further study with control/comparison groups may elucidate these differences further

The results supported the hypothesis that stigma is associated with higher frequency of maladaptive coping with a large effect size. Having a sense of feeling devalued by society through either direct experience of stigma, or the internalising of others' attitudes, can lead to an increased use of maladaptive coping strategies with the aim of alleviating levels of distress (44,51). Despite stigma being associated with higher levels of maladaptive coping, it was not found to be associated with lower levels of adaptive coping which does not support previous research relating to the relationship between stigma and coping strategies (89). However, a study examining HIV related stigma and distress also found similar results in that experienced stigma was correlated with maladaptive, but not adaptive coping (90). Additionally research looking at the mental health of students found that internalised stigma was also not correlated with adaptive coping (51). It may be the case that, although stigma may increase the likelihood of adopting

maladaptive coping strategies, this does not mean that it will decrease the use of any existing adaptive coping strategies.

As hypothesised, and in line with previous research in dystonia, stigma was associated with lower HRQOL (7). Stigma was also associated with increased psychological distress which is in line with previous research into Parkinson's disease, where higher levels of stigma were related to increased levels of distress (22,23,91). It has also been found that higher levels of stigma are related to lower levels of wellbeing in those with head, neck and lung cancer (92).

Maladaptive coping was also found to be associated with increased levels of distress, lower HRQOL and lower levels of wellbeing. This echoes previous research which has looked at maladaptive coping in relation to distress in dystonia (15,16,20,21), although the HRQOL and wellbeing findings specifically are novel.

Adaptive coping was not found to be significantly associated with measures of psychological distress (stress, anxiety or depression) or HRQOL. It was however, associated with the specific measure of wellbeing. A meta-analysis on coping strategies to regulate emotions found that the association between adaptive coping and distress was weaker than the association between maladaptive coping and distress (93), suggesting that the different strategies are not equal in their contribution to, and maintenance of distress. Research into the concept of psychological wellbeing has identified that this is more than an absence of psychological distress (94). Despite there often being an inverse correlation between wellbeing and distress, these are often moderate at best (55,95,96). This suggests they are separate rather than opposite constructs. Further support for this comes from the evidence that wellbeing and distress have distinct biological features (97). This might be why the findings for adaptive coping were specific to

wellbeing and, whilst use of such strategies might not reduce distress, they may be an important consideration in improving overall wellbeing.

When examined through a psychological mediation framework, maladaptive coping was found to mediate the relationship between stigma and distress, wellbeing and HRQoL which is in line with previous research in other populations that have explored this model (51,90). However, adaptive coping was not found to mediate the relationship between stigma and wellbeing in any of the psychological mediation models examined. This is contrary to previous research which found that adaptive coping mediated the relationship between stigma and psychological wellbeing or distress in students (51), or that it played a role in reducing symptoms of depression (98). Thus, the results from the current study only partially support the theory that coping strategies are processes which impact on wellbeing when trying to manage stress relating to stigma (44,51).

Rinehart et al. (90) also found no evidence for the mediating role of adaptive coping between stigma and psychological distress in a sample with HIV. They suggested that adaptive coping may serve more as a moderator that dampens the adverse effects of stigma. Support for this idea was found in another study which showed that adaptive coping was a moderating effect between HIV related stigma and medication adherence (99). Additionally, positive reframing (one of the sub categories of coping in the scale used in this study) has been shown to be a protective factor when trying to manage the stress associated with stigma in a study looking at a CBT intervention (100), so there may be benefit in future research of considering the separate coping strategies within the scale as opposed to grouping based on the two-factor model. Rather than the type of coping, how and when strategies are used may be more important when considering the impact on distress (101).

The psychological mediation framework (44) which provided the theoretical basis for the mediation model in this study was developed in the context of sexual minority stigma. Although the model has been applied in other areas such as HIV (102) and mental health (51) this is the first time that it has been applied to a health condition with a visible difference. The framework has been considered, via a systematic review, in relation to people living with obesity, another visible difference that is highly stigmatised (103) but to date this has remained hypothetical and not yet empirically tested.

### **Clinical implications**

As indicated here and elsewhere (7,8,38) people with dystonia can experience high levels of stigma and psychological distress, highlighting the need to consider these difficulties when working clinically with this population. A review of the literature highlighted that there is very little evidence for psychological interventions in dystonia (104). CBT is often the model of choice when managing anxiety and depression in this population (11) and there is some evidence for its effectiveness (53) although care should be given when working with distress that is related to stigma as negative responses and scrutiny from others are a reality for many people living with dystonia (38).

The results suggest the coping strategies have an important role to play in the relationship between stigma and distress. Developing skills relating to coping have been shown to be beneficial to mental health (105). These skills can be modelled and developed within psychological therapy both within group formats (106) and, individually using approaches such as Acceptance and Commitment Therapy (ACT). An important focus of ACT is behaviour change processes and valued action (107) which could help facilitate the development of alternative coping strategies, as well as promoting wellbeing (108).

On a more systemic level, initiatives that target the reduction of stigma relating to this condition may also decrease distress, improve wellbeing and enhance HRQOL. Stigma towards those with CD has been evidenced in the research (39) and the findings in this study suggest that the experiences of stigma from others and psychological distress are interrelated. A review on interventions for reducing health related stigma suggested that multiple levels of involvement should be considered including within the community, with those around the individual and at government and structural levels (109). For people with CD this might include the involvement of experts by experience in the design of any policies, providing training to health professionals to increase awareness of this condition and increasing public awareness.

### **Strengths and limitations**

The strengths of using a cross-sectional online survey methodology included an ease of access for participants, a minimisation of missing data and a geographically diverse sample. However, there are also some limitations. The majority of respondents were White British and female. Previous research has suggested a higher rate of females than males living with CD ranging from 61%-75% (110–112) though this is still considerably lower than the 82% of females in the current study which could affect generalisability across genders. Ethnicity is less frequently reported in studies on CD but in one study examining 66 people with CD they estimated the prevalence of white people with the condition to be 1.23 per 100,000 people and other races to be 0.15 per 100,000 people (113).

Although the option to receive the survey in paper form was available to participants, the advertisement was primarily online which may have excluded people who were unable to access or use computer technology. This could introduce a risk of bias to the study as younger people are more likely to take part in online research (114), although the mean age (52.3 years) was

similar to another UK study which recruited from an epidemiological study in the north of England (56.1 years) (17).

Categorising the concept of coping into two factors also has its limitations as the complexities and nuances of coping strategies can remain overlooked (48). A way to capture these would be to look at each strategy individually. However, as one of the purposes was to test the theoretical model it was more practical to use a simplified categorisation.

Hayes (78) discussed limitations in how findings from cross-sectional mediation are interpreted, as the direction of any relationships cannot be inferred. Therefore, although there were significant findings in this study, it cannot be assumed that stigma causes people with CD to cope with the stress in unhelpful ways which in turn causes them to have lower levels of psychological wellbeing. Rather, it appears there are interactions between the conceptualisations of stigma, coping and wellbeing, and it may be the case that some of these are bi-directional or that there are other factors also influencing these relationships.

### **Research Implications**

Given the limited research into the impact of stigma for people living with dystonia, and this being identified as an important aspect of their experience (40), it would be useful to build on the initial findings within this study to further explore the relationship between stigma and psychological wellbeing. This may be done through looking at the subscales of enacted and perceived stigma as well as examining in more detail any potential protective role of adaptive coping strategies. In addition, research into clinical interventions surrounding coping and stigma would be beneficial for this population.

As mentioned, there are some limitations to categorising coping into two factors. It would therefore be useful to further explore the experiences of coping in those with dystonia utilising a

qualitative methodology. This could then help inform decisions on the categorisation of coping strategies within quantitative research. A longitudinal methodology would be useful to explore any temporal and/or causal links between stigma and wellbeing in dystonia. It would also be useful to replicate this study with a more diverse population sample. This could be done by recruiting from existing epidemiological samples or via movement disorder clinics.

### **Conclusions**

The current study has provided valuable insights into the role of stigma and coping strategies on psychological wellbeing and HRQOL in CD. It is also the first study to apply the psychological mediation framework (44) in the context of a physical health condition with a visible difference. Levels of stigma were related to more frequent use of maladaptive coping, lower wellbeing, lower HRQOL and higher levels of distress. Maladaptive coping was related to lower levels of wellbeing, lower HRQOL and higher levels of distress. Adaptive coping was not related to stigma or distress, but more frequent use of adaptive coping was related to higher levels of wellbeing. In all the mediation models, maladaptive coping mediated the relationship between stigma, HRQOL and wellbeing. Adaptive coping did not mediate the relationship between stigma and any of the outcomes. Based on these findings and those in the wider literature, it is important to address coping strategies and stigma within clinical interventions and future research.

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Table 2-1

## Sample Demographics

Table 2-1

Sample demographics (n=114)

	n	%	Mean	Standard Deviation	Range
Age (years)			52.39	10.01	24-77
Age of symptom onset			37.49	12.86	9-67
Age of diagnosis			42.60	11.89	10-69
Gender:					
<i>Male</i>	21	18			
<i>Female</i>	93	82			
Ethnicity:					
<i>White English</i>	70	61			
<i>White Welsh</i>	5	4			
<i>White Scottish</i>	8	7			
<i>White British</i>	7	6			
<i>White Irish</i>	16	14			
<i>Other White Background</i>	5	4			
<i>Mixed White &amp; Black</i>	1	1			
<i>Caribbean</i>					
<i>Other</i>	2	2			
Partnership status:					
<i>Single</i>	22	19			
<i>Married/have partner</i>	83	73			
<i>Widowed</i>	3	3			
<i>Other</i>	6	5			
Living arrangements:					
<i>Alone</i>	17	15			
<i>With others</i>	94	83			
<i>Other</i>	2	2			
Work Status:					
<i>Employed full time</i>	29	25			
<i>Employed part time</i>	28	25			
<i>Not working</i>	38	33			
<i>Retired</i>	19	17			
FDQ scores (n=111):					
<42	45	40			
>43	67	60			

*Note:* Percentages are rounded to the nearest whole number, except for percentages less than one, which are rounded to the nearest 0.5%. FDQ = Functional Disability Questionnaire. Scores above the median of 42 indicate a moderate to severe level of disability relating to dystonia

**Table 2-2****Means, SDs and Cronbach's alpha of psychometric measures**

Table 2-2

Means, SDs and Cronbach's alpha of psychometric measures

Measure	M(SD)	Sample range	Cronbach's alpha
SSCI	61.33 (19.43)	25-115	0.95
COPE-M	22.58 (6.97)	12-43	0.81
COPE-A	36.41 (9.26)	16-57	0.84
CDIP-58	174.56 (54.17)	67-286	0.98
WEMWBS	41.68 (11.97)	14-70	0.95
DASS-S	8.60 (5.42)	0-21	0.89
DASS-A	6.69 (5.57)	0-21	0.87
DASS-D	8.60 (6.30)	0-21	0.93

*Note:* SSCI, Stigma Scale for Chronic Illness; COPE-M, COPE/Maladaptive subscale; COPE-A, COPE/Adaptive subscale; CDIP-58, Cervical Dystonia Impact Profile; WEMWBS, Warwick-Edinburgh Mental Wellbeing Scale; DASS21-S, Depression Anxiety Stress Scale/Stress subscale; DASS21-A, Depression Anxiety Stress Scale/Anxiety subscale; DASS21-D, Depression Anxiety Stress Scale/Depression subscale. Higher scores mean a higher level of what is being measured on each of the scales apart from the CDIP-58 where a higher score indicated a lower HRQOL.

**Table 2-3****Cut offs and frequency of severity of distress as measured using the DASS-21**

Table 2-3

Cut offs and frequency of severity of distress as measured using the DASS-21 – score (n; %)

	Depression	Anxiety	Stress
Normal	0-9 (50; 43.9%)	0-7 (47; 41.2%)	0-14 (53; 46.5%)
Mild	10-13 (9; 7.9%)	8-9 (4; 3.5%)	15-18 (19; 16.7%)
Moderate	14-20 (24; 21.1%)	10-14 (21; 18.4%)	19-25 (17; 14.9%)
Severe	21-27 (10; 8.8%)	15-19 (9; 7.9%)	26-33 (14; 12.3%)
Extremely Severe	28+ (21; 18.4%)	20+ (33; 30.0%)	34+ (11; 9.7%)
Total moderate – extremely severe	14+ (55; 48.3%)	10+ (63; 55.3%)	19+ (42; 36.8%)

*Note:* Calculated using the cut-offs proposed by (68) for the DASS. For DASS-21 scores are multiplied by two to calculate final score. Percentages rounded to one decimal place.

Table 2-4

## Non-parametric bivariate correlations among variables

Table 2-4

Non-parametric bivariate correlations among variables

	Age	Gender	SSCI	COPE-M	COPE-A	CDIP-58	WEMWBS	DASS21-S	DASS21-A	DASS21-D
Age	1	-0.17	-.183	-.158	-.030	.019	.151	-.094	-.030	-.073
Gender		1	-.016	.107	-.064	.219*	-.078	.133	.198*	.142
SSCI			1	.637**	.041	.641**	-.488**	.529**	.584**	.589**
COPE-M				1	.154	.528**	-.427**	.559**	.538**	.631**
COPE-A					1	-.068	.331**	.019	-.003	-.145
CDIP-58						1	-.707**	.686**	.670**	.729**
WEMWBS							1	-.539**	-.511**	-.669**
DASS21-S								1	.757**	.692**
DASS21-A									1	.740**
DASS21-D										1

*Note:* SSCI, Stigma Scale for Chronic Illness; COPE-M, COPE/Maladaptive subscale; COPE-A, COPE/Adaptive subscale;

CDIP-58, Cervical Dystonia Impact Profile; WEMWBS, Warwick-Edinburgh Mental Wellbeing Scale; DASS21-S, Depression

Anxiety Stress Scale/Stress subscale; DASS21-A, Depression Anxiety Stress Scale/Anxiety subscale; DASS21-D, Depression Anxiety

Stress Scale/Depression subscale.

\* $p < 0.05$

\*\* $p < 0.01$

**Table 2-5**

**Parallel Mediation Models**

Table 2-5

Parallel Mediation Models

	Model 1 X = stigma M <sub>1</sub> = adaptive coping M <sub>2</sub> = maladaptive coping Y = HRQoL	Model 2 X = stigma M <sub>1</sub> = adaptive coping M <sub>2</sub> = maladaptive coping Y = wellbeing	Model 3 X = stigma M <sub>1</sub> = adaptive coping M <sub>2</sub> = maladaptive coping Y = stress	Model 4 X = stigma M <sub>1</sub> = adaptive coping M <sub>2</sub> = maladaptive coping Y = anxiety	Model 5 X = stigma M <sub>1</sub> = adaptive coping M <sub>2</sub> = maladaptive coping Y = depression
a <sub>1</sub>	.003	.003	.003	.003	.003
a <sub>2</sub>	.246***	.246***	.246***	.246***	.246***
b <sub>1</sub>	-.456	.372***	-.020	-.030	-.135**
b <sub>2</sub>	2.006**	-.549**	.371***	.308***	.489***
c'	1.367***	-.194**	.061*	.091**	.091**
c	1.859***	-.328***	.152***	.167***	.210***
a <sub>1</sub> b <sub>1</sub>	-.001	.001	<.001	<-.001	<-.001
CI	-.060, .074	-.037, .038	-.004, .005	-.005, .005	-.016, .014
a <sub>2</sub> b <sub>2</sub>	.492 <sup>^</sup>	-.135 <sup>^</sup>	.091 <sup>^</sup>	.076 <sup>^</sup>	.120 <sup>^</sup>
CI	.136, .857	-.231, -.050	.050, .137	.035, .116	.075, .172
CSIE <sup>1</sup>	-.001	.002	<-.001	<-.001	-.001
CI	-.021, .026	-.062, .060	-.015, .019	-.018, .017	-.047, .041
CSIE <sup>2</sup>	.177 <sup>^</sup>	-.219 <sup>^</sup>	.327 <sup>^</sup>	.264 <sup>^</sup>	.370 <sup>^</sup>
CI	.051, .306	-.369, -.084	.184, .477	.122, .400	.237, .517

\*p < 0.05, \*\*p < 0.01, \*\*\*p < 0.001

<sup>^</sup> Significant indirect effect with 95% CI

<sup>1</sup> = adaptive coping as mediator

<sup>2</sup> = maladaptive coping as mediator

Note: X=predictor, M=mediator and Y=outcome; c' = direct effect of X on Y, controlling

for M; c = total effect of X on Y; ab = mediated effect; CI = confidence interval; CSIE:

completely standardised indirect effect.

**Figure 2-1**  
**Theoretical model**

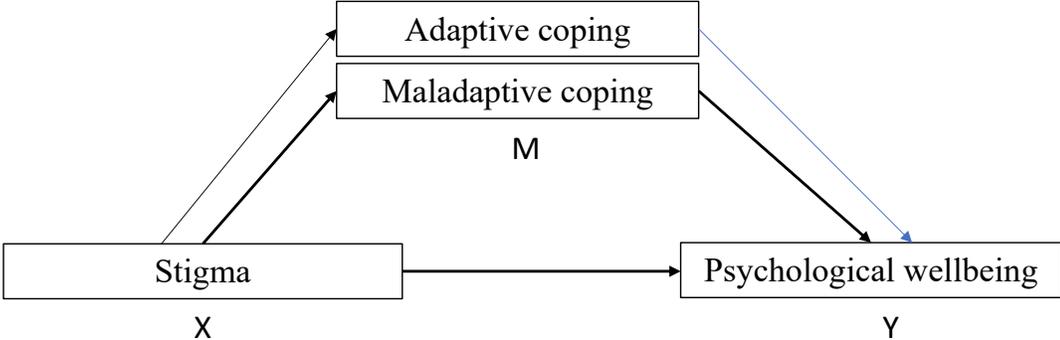
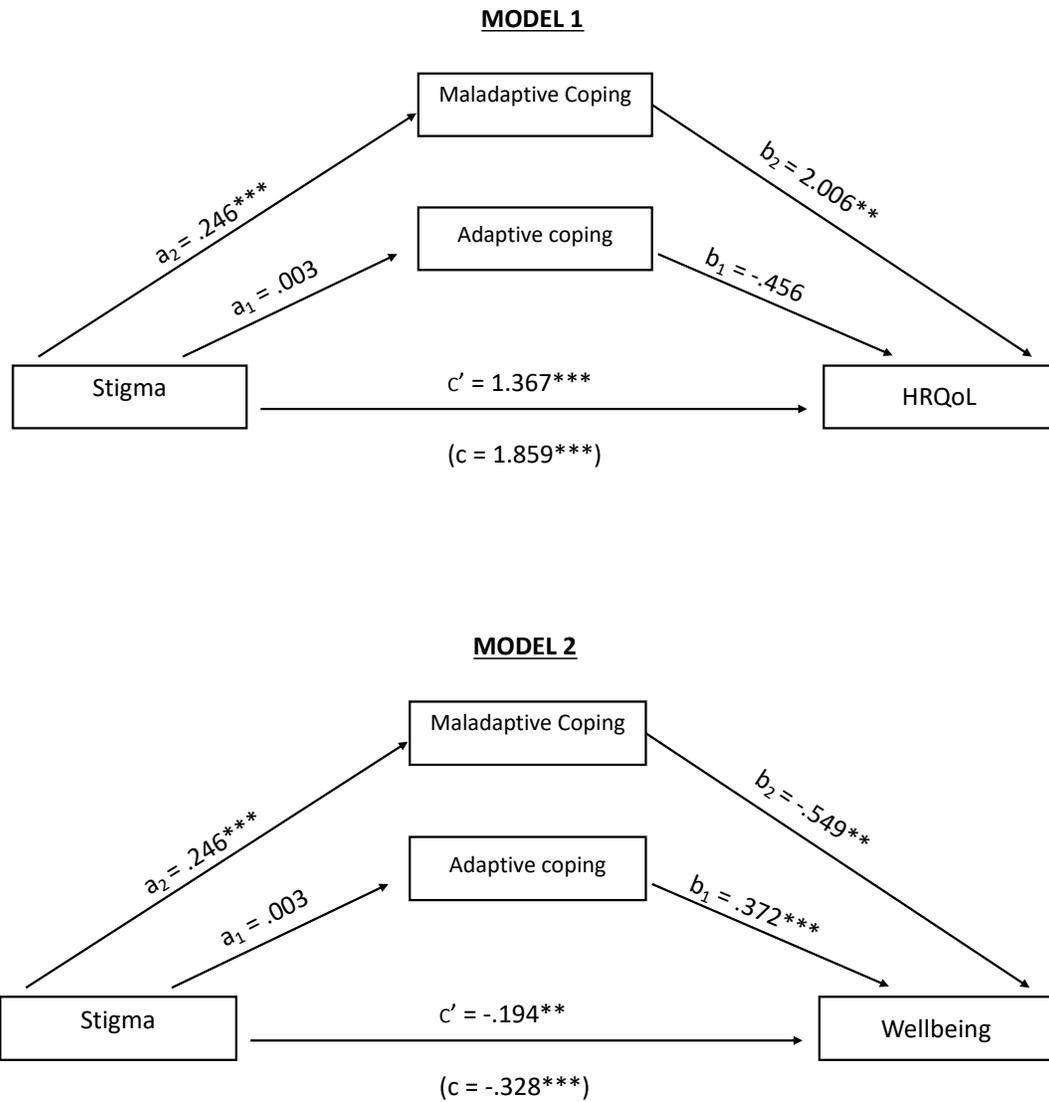
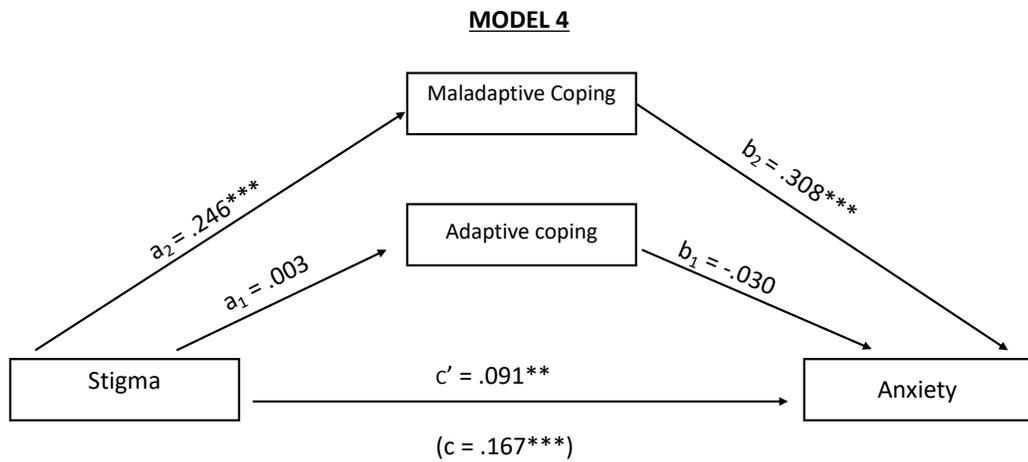
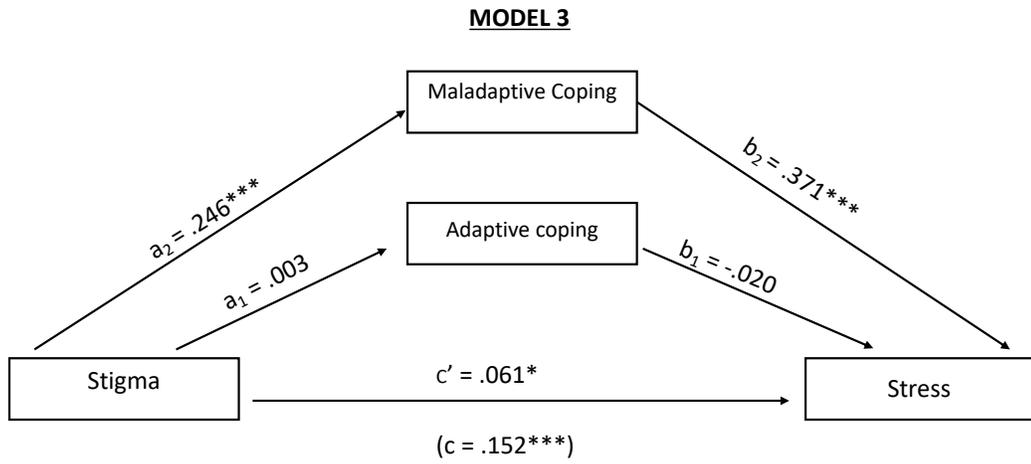


Figure 2-1: Theoretical model

Figure 2-2

Parallel Mediation Models





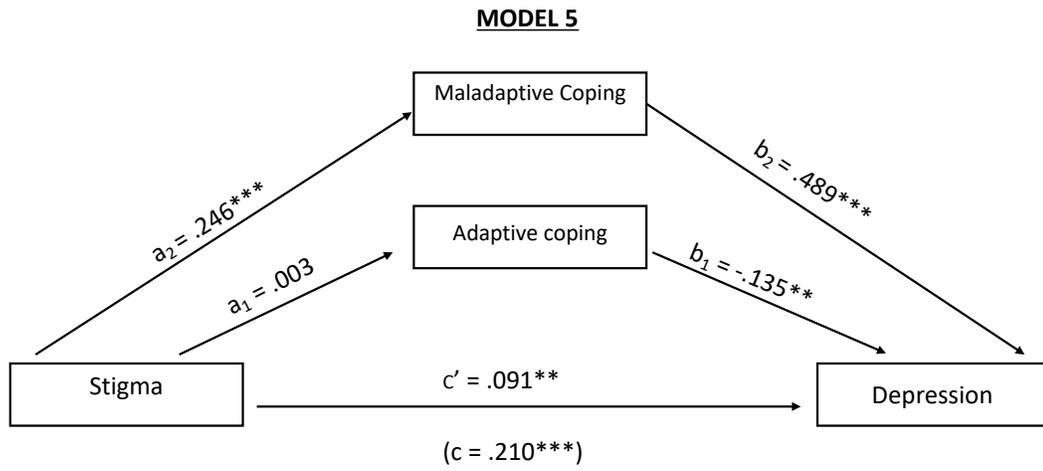


Figure 2-2. Parallel Mediation models (unstandardized).

Note: \*p < 0.05, \*\*p < 0.01, \*\*\*p < 0.001

## Appendix 2-1

### Author guidelines for *Psychology and Health*

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### **Section Three: Critical Appraisal**

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Helen Gowling

Doctorate in Clinical Psychology

Division of Health Research, Lancaster University

**All correspondence should be sent to:**

Helen Gowling  
Doctorate in Clinical Psychology  
Lancaster University  
Furness College  
United Kingdom  
LA1 4GY

Email : [h.gowling@lancaster.ac.uk](mailto:h.gowling@lancaster.ac.uk)  
Tel: 01524 59297

This critical appraisal will start by summarising the main findings of both the literature review and the research paper. It will also highlight the novel contribution of this research to the field of knowledge regarding psychological distress in dystonia. Following the summary, it will reflect on some of the decisions that were made throughout the project as well as considering the rationale for some of this. Within this is makes some suggestions for future research and highlights some of the ways the current project could have been improved on.

### **Main Findings**

#### ***Systematic literature review***

The systematic literature review examined the relationship between anxiety and depression and other psychological factors for those living with dystonia. Results were synthesized from 16 quantitative research papers. The findings suggested that psychological factors such as body image, coping styles, self-efficacy, personality traits and self-esteem were correlated with anxiety and/or depression in this population. There was also emerging evidence in the form of single studies which suggested that substance abuse, locus of control, sexual wellbeing and social support may also be correlated with anxiety and/or depression for this population. Overall there was a limited amount of research that had focussed on psychological factors relating to distress in dystonia, but it was clear that these were worthy of more consideration in future research and highlighted the need to do so. This also had clinical implications for the management of distress when working with this population.

#### ***Main paper***

The main paper was underpinned by a theoretical ‘psychological mediation framework’ model which proposes that the impact on psychological wellbeing from the stress related to stigma is mediated by the strategies a person uses to cope with such stress (1). This framework

was examined through the use of parallel mediational models with stigma as a predictor and with adaptive and maladaptive coping strategies as the mediator variables. Outcome variables were anxiety, depression, stress, health related quality of life (HRQOL) and wellbeing. A series of correlational analyses were also conducted to examine any potential relationships between the variables.

The results of the analysis suggested that higher levels of stigma were related to higher levels of distress, lower levels of wellbeing and lower HRQOL for people living with cervical dystonia. High levels of stigma were also related to more frequent use of maladaptive coping strategies but not to the use of adaptive coping strategies. A higher frequency of using maladaptive coping strategies was related to increased levels of distress, lower levels of wellbeing and lower HRQOL. A higher frequency of adaptive coping strategies was only found to be related to an increase in wellbeing. The mediation analysis identified an indirect effect of stigma on all the outcomes through maladaptive coping. Adaptive coping did not mediate the relationship between stigma and any of the outcomes. These findings provided partial support for the psychological mediation framework.

One of the strengths of this research was that it produced some novel findings that can be added to the developing understanding of the psychological impact of living with cervical dystonia, as well as support for the theoretical psychological mediation framework (1). Although this model has been examined in the context of sexual and gender minorities (2), mental health (3) and HIV (4), this study was the first time that it had been applied to those with a physical health condition which involves a visible difference. In addition, although stigma has previously been examined in relation to HRQOL (5), this is the first study to directly examine the

relationship between stigma and distress and wellbeing in this population. It is also the first to examine wellbeing in relation to other psychological factors in those with CD.

Clinical implications for thorough psychological formulations for people living with dystonia were identified, particularly due to the high reports of stigma in this cohort and the findings suggesting that maladaptive coping strategies may be related to the distress people experience as a result of being stigmatized. However, it was also discussed how adopting a more nuanced approach to the concept of coping strategies may help to further understand the complexities of their role in the development and maintenance of distress. This is discussed in more detail below but may include alternative categorisations of coping or qualitative research to further explore individual's experience of coping with the stress of stigma. The high rates of stigma reported in this study also highlight the importance of addressing societal attitudes on a more systemic level. Particularly as other research has highlighted that others show stigmatising attitudes to those with dystonia (6).

### **Decision-making, challenges and opportunities for improvement**

Throughout this piece of work there were many points where decisions had to be made that could have influenced its direction. I will therefore reflect on some of these below.

#### ***Systematic literature review***

##### ***Research question***

Attempting to formulate a research question for the systematic review was initially challenging. Research into dystonia has a strong medical focus and I was keen to bring my own interest of the role of clinical psychology in neurological conditions to this area. The challenge came from defining a question which was narrow enough to be able to effectively synthesise the

results from multiple papers, whilst also being broad enough to account for the limited psychological research that has traditionally been carried out with this population. An examination of the history of our understanding of dystonia suggests that the focus of psychological difficulties as a 'cause' of the condition has thankfully been gradually abandoned in favour of a more neurophysiological understanding, despite the mechanisms still not being fully understood (7). However, this has meant that the psychological distress commonly experienced by those with dystonia is often viewed as resulting from changes in the brain (8). Consideration of other psychosocial factors involved in the development and maintenance of psychological distress, although examined more extensively in other neurological disorders (9), has not had as much attention in dystonia. I had hoped that looking at psychological correlates of anxiety and depression in dystonia might help to bring focus onto other factors which may be involved in the development and maintenance of distress for those living with the condition, which in turn can be considered within clinical practice.

I made the decision to focus the research on idiopathic dystonia (which has no clear cause) as this is the most common presentation (10). Even within this category, there are many different presentations and there is research suggesting that there are differences in the experience of distress based on the location of the dystonia (11). As this is an already complex area, I made the decision to exclude studies which focussed on those who experienced dystonia as a side effect of taking psychiatric medications, or those where dystonia was secondary to another condition such as traumatic brain injury, as I felt that these would add extra complexity to an already complex area which was beyond the scope of this review.

### ***Search strategy***

I believe that my decision to keep my search strategy broad to increase sensitivity, and opt instead to manually screen a higher number of articles, was the right one to make to maintain the balance between sensitivity and specificity (12). Keeping the focus of the search simple to include only terms relating to ‘dystonia’, ‘anxiety’ and ‘depression’ meant that studies which included other psychological factors could be captured, even if these were not the main focus of the research. Additionally, distress in dystonia is conceptualised more as a ‘psychiatric’ difficulty than a ‘psychological’ one, so narrowing the search further to specify psychological factors may have meant that studies that were relevant would have been missed. The use of diagnostic language for psychological distress does not necessarily fit with my own, and some others’, views on diagnostic labels (13). However, this being an under researched area in a predominantly medical field, meant that adopting these terms for this project would mean that I accessed the largest pool of potential studies for inclusion in the review. In addition, as dystonia is typically understood and researched under the medical model and publications are often in medical, rather than psychological journals, using databases that covered both of these areas increased the chances of relevant papers being found.

### ***Research paper***

#### ***Survey design and measures***

Early on in the development of my empirical study, decisions had to be made concerning the measurement of key variables and the design of the online survey. A number of factors needed to be considered for the measures such as length, availability, reliability and validity. One of the strengths of this study was the contribution of two people with lived experience of cervical dystonia. Having their input on the proposal and survey design was invaluable, they were able to

make suggestions to improve on the language used to describe each of the measures, making it clearer and more accessible to those filling out the surveys. There were suggestions made on the wording of specific items within measures which I was unable to action as this would have impacted on the overall reliability. However, I was able to feedback to both individuals regarding the changes that were and were not made and the reasons for doing this.

As was found in my literature review, there have been a number of different measures used to look at psychological distress in those living with dystonia, but these are all general measures which have not been validated for use with this population. The issue here was that there were no measures that had been specifically validated for use with dystonia. The Becks Depression Inventory (BDI) (14) was a measure that had been commonly used in the research, although I felt that this would add too much length to the overall questionnaire when used alongside the other measures. There was some consideration given to the Hospital Anxiety and Depression Scale (HADS) (15), particularly as this has been suggested as a suitable screening for anxiety and depression in Parkinson's disease, which also has a motor component (16). However, as the underlying theoretical model that the research question was based on had an emphasis on stigma as a stressor, I made the decision to use the short version of the Depression Anxiety Stress Scale-21 (DASS-21) as this also included a subscale measuring levels of stress (17,18). I assessed it as being user friendly and it had been used in other studies involving movement disorders (19,20). The limitation from using this measure is that it had not specifically been used in dystonia before, so it was difficult to draw comparisons with previous research. In addition, the anxiety subscale of the DASS-21 has been found to be problematic in people with Parkinson's disease, another movement disorder with some similar features (21). However, this was more related to tremor so might be less relevant to this population. As results were similar

across the different outcome measures in my study, this may not have been problematic in the same way it has been in Parkinson's disease.

As there has only been limited research to date into the experience of psychological distress for those living with cervical dystonia, future research will need to further explore which measures accurately capture anxiety and depression in this population. One option would be to use the DASS-21 alongside other validated measures, such as the HADS or BDI, which have been utilised more extensively in dystonia and would be able to serve as a comparison. This would be more appropriate if there were fewer variables/measures included in the research design. It would also be useful for a comprehensive review and critique of the available measures to be conducted in relation to dystonia as this has not been carried out to date.

HRQOL was included alongside distress and wellbeing as an outcome measure to provide a more holistic picture of 'psychological wellbeing'. This was one variable for which there were two measures specifically designed for those with cervical dystonia: the Cervical Dystonia Impact Profile (CDIP-58) (22) and the Craniocervical Dystonia Questionnaire (CDQ-24) (23). Out of these the CDQ-24 would have been more preferable from a length perspective. However, it specifically includes a stigma subscale which may have inflated any findings of a relationship between stigma and quality of life. The CDIP-58 was longer and did not have stigma as a subscale, although there were stigma related items within the psychosocial functioning scale. The decision to use the CDIP-58 was based on the increased level of sensitivity in this population (24). It has also been found to correlate well with comparable subscales of the Medical Outcomes Study Short-Form 36-Item Health Survey (SF-36) (25) (24).

The SF-36 is a more generic health status measurement and has been used to assess HRQOL in previous dystonia studies (26). It has been recommended that both disease-specific

and generic measures should be used (27), which allows for a more complete picture of the impact of a condition, as well as being able to draw comparisons across the research with other conditions. Due to the length of the survey the decision to use one rather than two quality of life measures was taken. It may be pertinent for future research to use both types of measures to further understand the impact of stigma and coping styles on HRQOL. If this is done, the SF-36 is a good option as a general measure. Care should be taken though, as the computation of summary scores into a physical component score (PCS) and mental component score (MCS) as proposed by the original authors of the measure may not be suitable for those with cervical dystonia. A factor analysis carried out on the SF-36 found that the subscales of 'social functioning' and 'vitality', traditionally included in the MCS, were more strongly related to the PCS in cervical dystonia (28). Another consideration when using this measure is that the 'role physical' and 'role emotional' subscales had large floor and ceiling effects which can risk any clinical change being underestimated (28). However, this adds strength to the proposal of using a disease specific measure alongside, which may be more sensitive to changes within a specific condition.

With hindsight, the use of the CDIP-58 is one of the decisions that I may reconsider if replicating this study. The main reason for this was the length of the measure and the overall time it added to the survey. Although I had involvement from two individuals in the design of the project who had found the survey length manageable, once I had collected the data, there were a number of people who had not completed all the measures. I was unable to gather feedback from participants due to the anonymity of the survey, however, I hypothesised that the length may have been a factor in non-completion. This impacted on the amount of data available for analysis and risked the results being more biased towards those who were more able. I would suggest that

using the CDQ-24 alongside a further analysis to explore any impact of the stigma scale on the overall results, may make the survey more accessible to people.

*The concept of coping and use of language*

Throughout the process of designing, conducting and writing up my research paper, I have had several decisions to make around the inclusion of the concept of coping, including how to measure it, how to classify it, and how to name it. Some of the decisions have been quite difficult and, I feel, guided more by the aims and intended audience of the research rather than necessarily being reflective of my own values and beliefs. Coping itself is a difficult construct to conceptualise, with many differing descriptions and ways of measuring it within the literature (29). My literature review identified that it had been examined in the context of dystonia previously (30–33) and one of the measures used was the Ways of Coping Checklist (34). However, this is a 66-item measure, and as mentioned, I needed to consider the overall length and accessibility of the survey. Instead I opted for the Brief Cope (35). This was a more manageable length of 28 items covering 14 different subscales and was a validated way of measuring state coping strategies. It had also been used in other movement disorders such as Parkinson's disease and multiple sclerosis (36,37). In addition, it had been used in another study that examined the psychological mediation framework, which was underpinning my research, albeit with a cohort of students rather than in physical health.

Once I had chosen my scale then I needed to make a decision on the categorisation of the subscales. The measure has mainly been categorised in two ways: a two-factor categorisation (adaptive and maladaptive) and a three-factor categorisation (emotion focussed, problem focussed and dysfunctional).

I originally considered the three-factor categorisation, although felt that separating emotion-focussed coping and problem-focussed coping may be problematic. It has been suggested in the coping research that problem focussed coping strategies may be more adaptive than emotion focussed strategies (38,39). However, if a condition is chronic and there is no cure, individuals may feel that they have little control over the stressor and turn towards emotion-focussed rather than problem-focussed coping strategies (40). It has also been highlighted that problem-focussed strategies are more appropriate for controllable stressors and emotion-focussed strategies are more adaptive if the stressor is outside of an individual's control (41). There is also the issue that emotion focussed and problem focussed coping are not conceptually clear and most ways of coping could potentially fit into both categories (29). Therefore, as the scope of this study was to test the theoretical model for the first time in a novel area, a broader conceptualisation of coping strategies within the two-factor model felt more practical. This is also in line with other research that has used the theoretical model and same coping measure within a student population (3).

There are limitations to the two-factor categorisation that need to be considered. For example, 'religious coping' is included within the adaptive coping category. However, there have been mixed findings in the research with some supporting it as an adaptive strategy (42) and others (including in dystonia research) finding that it is a maladaptive strategy (31). For future research it would be worth looking at the individual subscales when analysing their mediating role in the relationship between stigma and psychological wellbeing. Another option would be to draw on a strategy used by Jahanshahi (31). Prior to entering maladaptive and adaptive coping into a regression model, they checked the correlations between the individual subscales and grouped them based on their relationship to a measure of depression. Although this

categorisation may limit the generalisability to the wider population, it would give a clearer idea of what coping styles may be adaptive or maladaptive for the specific group being studied. This could also be explored through qualitative research, examining how those living with dystonia cope with any stress associated with stigmatising experiences. Enhancing understanding around these concepts could then inform how coping is examined and categorised within quantitative research.

Despite the complexities of classification of coping, there is some agreement the ‘maladaptive’ and ‘adaptive’ are appropriate second-order dimensions (43,44), although I found the term ‘maladaptive’ did not fit with my own understanding of coping strategies. Further reading around classifications of coping styles revealed other descriptors such as ‘problematic’ and ‘dysfunctional’ which posed the same issues for me. These terms felt pathologizing and blaming, whereas a more nuanced understanding of coping strategies would recognise that there are times that even ‘maladaptive’ strategies would be adaptive and serve a purpose, and that strategies are not necessarily used in isolation of each other (45). Upon reflection I made the difficult decision to keep the wording, as this is the language that is used within the coping literature (43) and has also been used previously when looking at coping in dystonia(31). However, I may reconsider this if I was to replicate the research or examine coping styles in the future. One option would be to consult with those with lived experience and to include their thoughts and feelings on the terms used to describe the different coping strategies.

### ***Conclusion***

This project highlighted the importance of considering psychological approaches for people living with dystonia. Research within this area appears to be limited and with a focus on pathophysiological understandings of distress associated with this condition. The current review

of the literature and research paper revealed that there are other psychological factors involved with the distress experienced by those with dystonia. More research is needed employing a range of methodological designs (e.g. longitudinal, qualitative) to further understand the experiences of concepts such as stigma and coping, and how these factors interrelate in dystonia. An increase in our understanding of these will better enable clinicians to support the psychological wellbeing of people with dystonia.

Within this project there was numerous points where important decisions had to be made on how to proceed. One of the strengths of this was the involvement of people with lived experience of cervical dystonia and future research should strive to include this where feasible. When examining a number of different theoretical concepts (e.g. stigma, coping, quality of life) within a single research design, there will always be limitations due to the complexities of these areas and decisions have to be made to balance the number and length of measures against the accessibility for those agreeing to take part. On the whole I feel that most of the decisions I made were the right ones for this project. The main considerations I would give if repeating this research would be to use a shorter measure of disease specific quality of life, and maybe add a generic health impact measure such as the SF-36. I would also consider exploring different categorisations of coping strategies.

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**Section Four: Ethics Section**

Word Count (excluding references, tables and appendices):

Helen Gowling

Doctorate in Clinical Psychology

Division of Health Research, Lancaster University

**All correspondence should be sent to:**

Helen Gowling  
Doctorate in Clinical Psychology  
Lancaster University  
Furness College  
United Kingdom  
LA1 4GY

Email : [h.gowling@lancaster.ac.uk](mailto:h.gowling@lancaster.ac.uk)  
Tel: 01524 59297

## 1 Ethics Proposal

Faculty of Health and Medicine Research Ethics Committee (FHMREC)

Lancaster University

Application for Ethical Approval for Research

**for additional advice on completing this form, hover cursor over 'guidance'.**

**Guidance on completing this form is also available as a word document**

**Title of Project:** Stigma, coping styles and wellbeing in individuals with cervical dystonia

**Name of applicant/researcher:** Helen Gowling

**ACP ID number (if applicable)\*:**

**Funding source (if applicable)**

**Grant code (if applicable):**

**\*If your project has *not* been costed on ACP, you will also need to complete the Governance Checklist [\[link\]](#).**

### **Type of study**

- Involves existing documents/data only, or the evaluation of an existing project with no direct contact with human participants. **Complete sections one, two and four of this form**
- Includes *direct* involvement by human subjects. **Complete sections one, three and four of this form**

**SECTION ONE**

**1. Appointment/position held by applicant and Division within FHM** Trainee Clinical Psychologist,  
Division of Health and Medicine

**2. Contact information for applicant:**

**E-mail:** h.gowling@lancaster.ac.uk

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

**Telephone:** 07913203650

**Address:** Division of Health Research, Faculty of Health and Medicine, C37 - Furness College, Lancaster University, Lancaster, LA1 4YG

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

- i) Helen Gowling – Trainee Clinical Psychologist
- ii) Dr Fiona Eccles – Lecturer in health research
- iii) Dr Fiadhait O’Keeffe – Senior Clinical Neuropsychologist

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

PG Diploma  Masters by research  PhD Thesis  PhD Pall. Care

PhD Pub. Health  PhD Org. Health & Well Being  PhD Mental Health  MD

DClinPsy SRP  [if SRP Service Evaluation, please also indicate here:  DClinPsy Thesis

**4. Project supervisor(s), if different from applicant:** Dr Fiadhait O’Keeffe and Dr Fiona Eccles

**5. Appointment held by supervisor(s) and institution(s) where based (if applicable):**

Senior Clinical Neuropsychologist	Lecturer in Research Methods
St Vincent's University Hospital	Furness Building
Elm Park	Lancaster University
Dublin 4	Lancaster
DO4 T6F4	LA1 4YG

## SECTION TWO

**Complete this section if your project involves existing documents/data only, or the evaluation of an existing project with no direct contact with human participants**

1. Anticipated project dates (month and year)

Start date:

End date:

2. Please state the aims and objectives of the project (no more than 150 words, in lay-person's language):

### Data Management

*For additional guidance on data management, please go to [Research Data Management](#) webpage, or email the RDM support email: [rdm@lancaster.ac.uk](mailto:rdm@lancaster.ac.uk)*

3. Please describe briefly the data or records to be studied, or the evaluation to be undertaken.

4a. How will any data or records be obtained?

4b. Will you be gathering data from websites, discussion forums and on-line 'chat-rooms'

4c. If yes, where relevant has permission / agreement been secured from the website moderator?

4d. If you are only using those sites that are open access and do not require registration, have you made your intentions clear to other site users?

4e. If no, please give your reasons

5. What plans are in place for the storage, back-up, security and documentation of data (electronic, digital, paper, etc)? Note who will be responsible for deleting the data at the end of the storage period. Please ensure that your plans comply with General Data Protection Regulation (GDPR) and the (UK) Data Protection Act 2018.

6a. Is the secondary data you will be using in the public domain?

6b. If NO, please indicate the original purpose for which the data was collected, and comment on whether consent was gathered for additional later use of the data.

Please answer the following question *only* if you have not completed a Data Management Plan for an external funder

7a. How will you share and preserve the data underpinning your publications for at least 10 years e.g. PURE?

7b. Are there any restrictions on sharing your data?

### **8. Confidentiality and Anonymity**

a. Will you take the necessary steps to assure the anonymity of subjects, including in subsequent publications?

b. How will the confidentiality and anonymity of participants who provided the original data be maintained?

9. What are the plans for dissemination of findings from the research?

10. What other ethical considerations (if any), not previously noted on this application, do you think there are in the proposed study? How will these issues be addressed?

### SECTION THREE

**Complete this section if your project includes *direct* involvement by human subjects**

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

This study is designed to look at how the stress of stigma, and strategies that somebody uses to cope with that stress, might impact on overall psychological well-being and quality of life for people diagnosed with cervical dystonia, a condition which affects the muscles in the head and neck.

This will be examined through surveys designed to explore stigma, coping styles and wellbeing. Participants for the study will be recruited via two dystonia specific charities and invited to complete the surveys either online or on paper. The data from these surveys will then be analysed to explore whether coping strategies impact on the relationship between the stress of stigma and wellbeing/quality of life.

2. **Anticipated project dates (month and year only)**

Start date: September 2019

End date: May 2020

#### **Data Collection and Management**

*For additional guidance on data management, please go to [Research Data Management](#) webpage, or email the RDM support email: [rdm@lancaster.ac.uk](mailto:rdm@lancaster.ac.uk)*

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

minimum 116 and maximum 300 participants of any gender

#### **Inclusion criteria**

- Individuals who self-report a diagnosis of cervical dystonia and are 18 and older.

- Individuals who will be able to complete the measures alone or with support.
- The questionnaire will be written in English, so individuals will need to have sufficient understanding of written English to take part.

**Exclusion criteria**

- Potential participants will be excluded if they have dystonia following a serious injury, or any other significant illness/condition that affects their visible appearance.

4. How will participants be recruited and from where? Be as specific as possible. Ensure that you provide the *full versions* of all recruitment materials you intend to use with this application (e.g. adverts, flyers, posters).

This project will be advertised through the Dystonia Society (<https://www.dystonia.org.uk/>) and Dystonia Ireland (<http://www.dystonia.ie/>) via any channels available to them (e.g. website, social media, newsletter, groups, posters) and using both a poster and a video. Other dystonia relevant online organisations may also be approached to request the advert be shared through their channels. It will also be advertised on the Lancaster University Doctorate in Clinical Psychology webpage and social media. An advert will be used to inform people of the research and invite them to take part. Individuals will read information about the purpose of the study and the length of time it should take (30 minutes) before being directed to a consent page if they wish to take part. Once consent has been provided to take part in the research the online survey will appear. If participants would prefer to complete the survey in paper format, the information page and initial advert will include contact details and instructions on how to access a paper copy.. Paper copies of the survey will be provided if participants choose to complete the survey in this format. At the end of the study participants will be debriefed and details of support resources as provided in the initial information will be repeated.

5. Briefly describe your data collection and analysis methods, and the rationale for their use.

Data will be collected via online survey tool, Qualtrics. Paper surveys will also be provided which will be manually entered into Qualtrics when they have been returned. The survey will comprise of demographic/ clinical details (age, gender, ethnicity, work status, relationship status, living arrangements, age at onset, duration, time since diagnosis, medication, treatment, other conditions) and the following validated measures:

- The short version of the Depression Anxiety Stress Scale – 21 (DASS-21) (Henry & Crawford, 2005; Lovibond & Lovibond, 1995). This is a 21-item scale. Although not used specifically with people living with dystonia, it has been used with other neurological conditions which present with movement disorders such as Parkinson’s disease (Birtwell, Dubrow-Marshall, Dubrow-Marshall, Duerden, & Dunn, 2017) and multiple sclerosis (Solaro, Gamberini, & Masuccio, 2018).

- The Warwick-Edinburgh Mental Well-being Scale (WEMWBS) (Tennant et al., 2007) is a 14-item validated measure of wellbeing which focusses on positive aspects of mental health. Although not researched specifically with physical health populations, it is considered easy to complete and useful for general population samples (Koushede et al., 2019).
- Quality of Life will be measured using the Cervical Dystonia Impact Profile (CDIP-58) (Cano et al., 2004) which is a 58-item scale measuring the impact of CD across eight domains: head and neck symptoms, pain and discomfort, upper limb activities, annoyance, sleep, walking, mood and psychosocial functioning. This measure has been shown to have good validity and reliability (Cano et al., 2008).
- Stigma will be measured by the 24-item Stigma Scale for Chronic Illness (SSCI). This scale was developed for people with chronic neurological disorders and measures both felt stigma and enacted stigma (Rao et al., 2009). It has been shown to have fair internal consistency and good content validity (Stevellink, Wu, Voorend, & van Brakel, 2012).
- Coping styles will be measured using the brief COPE (Carver, 1997). This has 14 scales with each subscale including two items. Although this has been categorised in various ways in the literature, this study will use the two-category model developed by Meyer (2001) which divides the subscales into adaptive and maladaptive coping strategies.
- Disease Severity will be measured using the Functional Disability Questionnaire (Jahanshahi & Marsden, 1990). This is a 27-item scale developed to measure the impact of dystonia on activities of daily living. The construct and concurrent validity of the scale, along with test-retest reliability and internal consistency were shown to be high (Jahanshahi & Marsden, 1990).

A series of mediation analyses using Hayes PROCESS tool (Hayes, 2013) will be used to examine whether coping style mediates the relationship between stigma and psychological outcomes.

6. What plan is in place for the storage, back-up, security and documentation of data (electronic, digital, paper, etc.)? Note who will be responsible for deleting the data at the end of the storage period. Please ensure that your plans comply with General Data Protection Regulation (GDPR) and the (UK) Data Protection Act 2018.

Any paper copies of the consent form and survey will be entered into the electronic data set upon receipt and the paper version immediately and securely destroyed in confidential waste. During the collection and analysis of the data, the electronic data set will be stored in a password protected file in the University's H drive. Once the project has been examined the electronic data and electronic consent forms will be stored securely in a password protected file by the Doctorate in Clinical Psychology Course at Lancaster University for 10 years. These will be accessible by the Research Coordinator and Fiona Eccles (Research supervisor) who will be the data custodian. After 10 years the Research Coordinator will destroy the data under instruction from the research supervisor.

Any contact details provided by participants who wish to receive a copy of the final report will be stored in a password protected and encrypted file in the University's H drive until the project is complete and the report has been sent out. At this point this file will be securely destroyed.

7. Will audio or video recording take place?  no  audio  video

a. Please confirm that portable devices (laptop, USB drive etc) will be encrypted where they are used for identifiable data. If it is not possible to encrypt your portable devices, please comment on the steps you will take to protect the data.

b. What arrangements have been made for audio/video data storage? At what point in the research will tapes/digital recordings/files be destroyed?

Please answer the following questions *only* if you have not completed a Data Management Plan for an external funder

8a. How will you share and preserve the data underpinning your publications for at least 10 years e.g. PURE?

The raw data will not be shared. Full data will only be accessible to Helen Gowling (lead researcher) Fiona Eccles (research supervisor), Fiadhnaid O'Keefe (field supervisor) and the Research Coordinator for Lancaster University's Doctorate in Clinical Psychology. After 10 years the Research Coordinator will be responsible for destroying the data under instruction from the research supervisor.

8b. Are there any restrictions on sharing your data?

Raw data will be kept confidential and only accessed by the research team. However, data may be shared with other genuine researchers if requested once the project is complete, but only in an aggregated form (i.e. age groups rather than individual ages).

9. Consent

a. Will you take all necessary steps to obtain the voluntary and informed consent of the prospective participant(s) or, in the case of individual(s) not capable of giving informed consent, the permission of a legally authorised representative in accordance with applicable law?  yes

b. Detail the procedure you will use for obtaining consent?

Individuals will read information about the purpose of the study before being directed to a consent page if they wish to take part. Once consent has been provided to take part in the research the online survey will appear. If participants would prefer to complete the survey in paper format, the information page

and original advertisement will include contact details and instructions on how to access a paper copy. Paper copies of the survey will be sent including information sheet and consent forms.

10. What discomfort (including psychological e.g. distressing or sensitive topics), inconvenience or danger could be caused by participation in the project? Please indicate plans to address these potential risks. State the timescales within which participants may withdraw from the study, noting your reasons.

Participating in this study should not pose a significant risk to participants. However, reflecting on the issues raised within the research may be difficult for some people. Contact details will be provided of places that can be provide support should any issues arise. Additionally, it will be made clear prior to starting the study that they can stop at any time during the survey although, due to the anonymity of participation, their data cannot be removed after they have agreed to take part.

11. 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).

There are no anticipated risks for the researcher. However, if any issues should arise I will seek support and supervision from Fiona Eccles (research supervisor).

12. 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.

There are no direct benefits.

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

For participants who would prefer to access the survey in paper format, an additional cost of postage will be incurred. This will be funded by the DClinPsy course at Lancaster in the form of pre-paid postage.

14. Confidentiality and Anonymity

a. Will you take the necessary steps to assure the anonymity of subjects, including in subsequent publications?  yes

b. Please include details of how the confidentiality and anonymity of participants will be ensured, and the limits to confidentiality.

There will be nothing within the survey that would link the responses to any individual.

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

Two experts by experience have been consulted on the measures used within the survey and the design of the project.

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

Findings will be shared with the Dystonia Society and Dystonia Ireland to share with their members. In addition, a report detailing the findings will be sent to participants that have requested to receive this once the project is complete. The findings will contribute to the lead researcher's thesis which will be presented at a thesis presentation day at Lancaster University. If the findings are appropriate they may then also be shared in conferences, special interest groups or a formal journal publication

17. 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?

**SECTION FOUR: signature**

Applicant electronic signature: Helen Gowling

Date 09.08.19

Student applicants: please tick to confirm that your supervisor has reviewed your application, and that they are happy for the application to proceed to ethical review

Project Supervisor name (if applicable): Fiona Eccles

Date application discussed 2.7.19

**Submission Guidance**

1. **Submit your FHMREC application by email to Diane Hopkins**  
([fhmresearchsupport@lancaster.ac.uk](mailto:fhmresearchsupport@lancaster.ac.uk)) as two separate documents:
  - i. **FHMREC application form.**  
Before submitting, ensure all guidance comments are hidden by going into 'Review' in the menu above then choosing *show markup>balloons>show all revisions in line*.
  - ii. **Supporting materials.**  
Collate the **following materials for your study, if relevant, into a single word document:**
    - a. **Your full research proposal (background, literature review, methodology/methods, ethical considerations).**
    - b. Advertising materials (posters, e-mails)
    - c. Letters/emails of invitation to participate
    - d. Participant information sheets
    - e. Consent forms
    - f. Questionnaires, surveys, demographic sheets
    - g. Interview schedules, interview question guides, focus group scripts
    - h. Debriefing sheets, resource lists

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

2. Submission deadlines:
  - i. Projects including direct involvement of human subjects [**section 3 of the form was completed**]. The *electronic* version of your application should be submitted to [Becky Case](#) by the committee deadline date. Committee meeting dates and application submission dates are listed on the [FHMREC website](#). Prior to the FHMREC meeting you

may be contacted by the lead reviewer for further clarification of your application. Please ensure you are available to attend the committee meeting (either in person or via telephone) on the day that your application is considered, if required to do so.

- ii. The following projects will normally be dealt with via chair's action, and may be submitted at any time. **[Section 3 of the form has *not* been completed, and is not required]**. Those involving:
    - a. existing documents/data only;
    - b. the evaluation of an existing project with no direct contact with human participants;
    - c. service evaluations.
3. **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**

## 2 Study Protocol

### **Stigma, coping styles and wellbeing in individuals with cervical dystonia.**

V

#### **Applicants**

##### *Principal Investigator*

Helen Gowling

Trainee Clinical Psychologist, Doctoral Student in Clinical Psychology (DClinPsy)

Room C34, Furness College, Lancaster University, Lancaster, LA1 4YG

Tel: 44 (0)7913203650, email: h.gowling@lancaster.ac.uk

##### *Research Supervisor*

Dr Fiona Eccles

Lecturer in health research, Lancaster University, Lancaster, LA1 4YT, UK

Tel: 44 (0)1524 592807, email: f.eccles@lancaster.ac.uk

##### *Field Supervisor*

Dr Fiadhnaith O’Keeffe

Senior Clinical Neuropsychologist, St. Vincent’s University Hospital, Elm Park, Dublin  
4, D04 T6F4

Email: fokeeffe@svuh.ie

#### **Introduction**

Dystonia is a movement disorder which involves repetitive movements and abnormal postures caused by sustained muscle contractions (Lewis, Butler, & Jahanshahi, 2008). Studies have shown that it affects approximately 0.01-0.03% of the population (Phukan, Albanese, Gasser, & Warner, 2011) The effects of the visible changes to appearance have been associated with embarrassment which can lead to psychological difficulties, social avoidance, and isolation (Lewis et al., 2008). Cervical dystonia, also known as spasmodic torticollis, is the most common kind of focal dystonia (a type of dystonia that affects one part of the body) and is characterised by uncontrolled twisting, turning and tilting of the head caused by involuntary and often painful contractions of the neck muscles (Ben-Shlomo, Camfield, & Warner, 2002).

One focus of research in this population has been health related quality of life (HRQOL) (Weiss et al., 2017). This broadly looks at how a condition impacts on an individual across physical, social and psychological domains (Calvert & Freemantle, 2003; Hays, Hahn, & Marshall, 2002). The research suggests that depression (Basurović, Svetel, Pekmezović, & Kostić, 2012), negative body concept (Pekmezovic et al., 2009) and social participation (Ben-Shlomo et al., 2002) are some of the main factors correlated with HRQOL. In studies of other movement disorders, such as Parkinson's Disease, stigma has also been found to play a key role in HRQOL (Ma, Saint-Hilaire, Thomas, Tickle-Degnen, & Thomas, 2016)

Stigma is a vital aspect of health research. Visible characteristics, such as tremors or muscle contractions in dystonia, can result in stigmatising social experiences alongside the physical and emotional impact of the condition (Joachim & Acorn, 2000). Psychological, physical and social aspects of illness can be exacerbated by the emotional impact of social disqualification (Weiss, Ramakrishna, & Somma, 2006). Stigma relating to a condition may contribute towards a decrease in overall quality of life (Molina, Choi, Cella, & Rao, 2013).

Stigma can be broken down into two main areas: enacted stigma which is related to discrimination and attitudes from the public; and perceived stigma which is an internalising of enacted stigma by an individual, often leading to feelings of shame (Goffman, 1963). Perceived stigma has also been shown to lead to a poorer quality of life, negatively impact health related outcomes and increase social isolation in people with mental health difficulties (Fung, Tsang, & Corrigan, 2008; Rüsç & Corrigan, 2010; Rüsç et al., 2006). Studies of enacted stigma with populations of people with epilepsy show that frequency of enacted stigma is associated with increased psychological distress and a poorer quality of life (Kumari, Ram, Haque Nizamie, & Goyal, 2009; McLaughlin, Pachana, & McFarland, 2008; Suurmeijer, Reuvekamp, & Aldenkamp, 2001). Stigma has also been found to predict psychological outcomes in people with neurological conditions which present with movement disorders such as Parkinson's disease and multiple sclerosis (Gallagher, Lees, & Schrag, 2010; Valvano et al., 2016).

The role of stigma in psychological outcomes for people with dystonia is poorly understood. One study found a significant relationship between stigma and both the mental and physical aspects of quality of life in people with cervical dystonia (Ben-Shlomo et al., 2002). Another study found no significant relationship between stigma and health related quality of life for people with segmental dystonia (Basurović et al., 2012), however this study had a small sample size. Both studies also used a stigma scale designed for people with bowel cancer (MacDonald & Anderson, 1984). A 2001 study examining the role of stigma in cervical dystonia found that the majority of participants had felt some, or severe stigma (Papathanasiou, MacDonald, Whurr, & Jahanshahi, 2001). The questionnaire used in the 2001 study was created following interviews with participants. Although this meant that it had good face validity and was focussed on what was important to the 10 participants initially interviewed, it is unclear how

generalisable this could be to other people living with dystonia. An additional study looking at the perceptions of people without dystonia showed stigmatising attitudes towards those with cranial and cervical dystonia (Rinnerthaler, Mueller, Weichbold, Wenning, & Poewe, 2006), suggesting that enacted stigma is very much a part of daily life for this population, particularly those with cervical dystonia.

Research focussed on other groups who experience stigma draws heavily on the minority stress paradigm (Meyer, 2003), which conceptualises stigma as a stressor above and beyond what non-stigmatised people endure, which then leads to difficulties with mental health. This was further developed into a psychological mediation framework (Hatzenbuehler, 2009) which integrated the theory of minority stress with psychological processes that may result from being stigmatised, and therefore contribute to psychological distress. One of these proposed processes was coping styles. There is emerging evidence that the experience of stigma can lead to coping behaviours which could be considered maladaptive, such as disengagement and substance use, which in turn can impact on psychological wellbeing and quality of life (Hatzenbuehler, Phelan, & Link, 2013). To date the relationship between stigma, coping styles and psychological outcomes has not been examined in people with dystonia.

Consequently, this study will investigate the relationship between stigma and psychological outcomes then also investigate whether coping mediates this relationship in a cohort of people with cervical dystonia. The focus will be on people with cervical dystonia as this is the most common type of dystonia. If stigma and coping are found to be a significant factor for distress and wellbeing, then this would have implications for the field of clinical psychology on both an individual and a systemic level, through potential psychological

interventions and, in turn, potentially lead to improved psychological well-being for those with cervical dystonia.

### **Method: Participants**

For a medium effect (0.36) in both paths (stigma to control ( $\alpha$ ) and control to psychological outcomes ( $\beta$ )) using a bias-corrected bootstrap mediation model, approximately 71 participants are required for 80% power, rising to 115-116 if the  $\alpha$  or  $\beta$  path coefficient has a small-medium effect (0.26) (Fritz & MacKinnon, 2007). Previous research looking at the relationship between stigma and coping have found small-medium effects for adaptive coping and large effects for maladaptive coping, suggesting powering the study for a small-medium effect should be sufficient (Tran & Lumley, 2019). It is not possible to take the effect size for  $\beta$  from previous research as this requires the identical model (and we are not aware of such research). However, the study will be powered so that it should find a small-medium effect should it exist (at 80% power). Thus, in total the aim would be to recruit 116 participants

### **Inclusion criteria**

- Individuals who self-report a diagnosis of cervical dystonia
- aged 18 or over.
- Individuals who are able to complete the measures alone or with support.
- The questionnaire will be written in English, so individuals will need to have sufficient understanding of written English to take part.

### **Exclusion criteria**

- Potential participants will be excluded if they have dystonia following a serious injury,
- Potential participants who also have other significant illness/condition that affects their visible appearance will also be excluded.

**Method: Design**

The study will be a cross-sectional survey using quantitative measures. The data will be quantitatively examined and a mediation analysis will be conducted using Hayes process tool (Hayes, 2013) within SPSS to examine whether coping style mediates the relationship between stigma and psychological wellbeing (including distress) and quality of life.

The dependent variables will be scores of emotional wellbeing, distress, and quality of life: The short version of the Depression Anxiety Stress Scale – 21 (DASS-21) (Henry & Crawford, 2005; Lovibond & Lovibond, 1995), the Warwick-Edinburgh Mental Well-being Scale (WEMWBS) (Tennant et al., 2007), and the Cervical Dystonia Impact Profile (CDIP-58) (Cano et al., 2004) which assess quality of life will be used. – see materials section for details on reliability and validity.

The predictor variable will be perceived stigma, measured using the Stigma Scale for Chronic Illness (SSCI-8) (Rao et al., 2009). The mediating variable will be coping style which will be measured using the brief COPE (Carver, 1997) using the two-category model developed by Meyer (2001) which divides the subscales into adaptive and maladaptive coping strategies.

**Method: Materials**

The survey will contain demographic, clinical and validated measures. (See appendices 3-9)

**Demographic variables**

- Age
- Gender
- Ethnicity

- Work status
- Relationship status
- Living arrangements

### **Clinical variables**

- Age of onset
- Duration
- Time since diagnosis
- Medication
- Botulinum toxin (BTX) treatment
- Other treatments
- Other health conditions

### **Validated measures**

- The short version of the Depression Anxiety Stress Scale – 21 (DASS-21) (Henry & Crawford, 2005; Lovibond & Lovibond, 1995). This is a 21-item scale. Although not used specifically with people living with dystonia, it has been used with other neurological conditions which present with movement disorders such as Parkinson's disease (Birtwell, Dubrow-Marshall, Dubrow-Marshall, Duerden, & Dunn, 2017) and multiple sclerosis (Solaro, Gamberini, & Masuccio, 2018).
- The Warwick-Edinburgh Mental Well-being Scale (WEMWBS) (Tennant et al., 2007) is a 14-item validated measure of wellbeing which focusses on positive aspects of mental health. Although not researched specifically with physical health populations, it is considered easy to complete and useful for general population samples (Koushede et al., 2019).

- Quality of Life will be measured using the Cervical Dystonia Impact Profile (CDIP-58) (Cano et al., 2004) which is a 58-item scale measuring the impact of CD across eight domains: head and neck symptoms, pain and discomfort, upper limb activities, annoyance, sleep, walking, mood and psychosocial functioning. This measure has been shown to have good validity and reliability (Cano et al., 2008).
- Stigma will be measured by the 24-item Stigma Scale for Chronic Illness (SSCI). This scale was developed for people with chronic neurological disorders and measures both felt stigma and enacted stigma (Rao et al., 2009). It has been shown to have fair internal consistency and good content validity (Stevelink, Wu, Voorend, & van Brakel, 2012).
- Coping styles will be measured using the brief COPE (Carver, 1997). This has 14 scales with each subscale including two items. Although this has been categorised in various ways in the literature, this study will use the two-category model developed by Meyer (2001) which divides the subscales into adaptive and maladaptive coping strategies.
- Disease Severity will be measured using the Functional Disability Questionnaire (Jahanshahi & Marsden, 1990). This is a 27-item scale developed to measure the impact of dystonia on activities of daily living. The construct and concurrent validity of the scale, along with test-retest reliability and internal consistency were shown to be high (Jahanshahi & Marsden, 1990).

### **Method: Procedure**

This project will be advertised through the UK Dystonia Society (<https://www.dystonia.org.uk/>) and Dystonia Ireland (<http://www.dystonia.ie/>) via any channels available to them (e.g. website, social media, newsletter, groups, posters) and using both an advert (appendix 12) and a video (appendix 13). The video will have the advert visible below it

on the webpage to provide people with the relevant contact information. Other dystonia relevant online organisations may also be approached to request the advert be shared through their channels. It will also be shared on the Lancaster University Doctorate in Clinical Psychology webpage and social media. An advert will be provided however the charities will be free to add their own wording when sharing details of the study with their members (see appendix 12 for advert). Individuals will read information about the purpose of the study before being directed to a consent page if they wish to take part (see appendices 1 and 2 for participant information and consent form- paper version). Once consent has been provided to take part in the research the online survey will appear (see appendix 11 for link to online version of the survey). If participants would prefer to complete the survey in paper format, the information page and initial advert will include contact details and instructions on how to access a paper copy. Paper copies of the survey will be provided if participants choose to complete the survey in this format. At the end of the study participants will be provided with a debrief, information on how they can be entered into a prize draw in return for taking part, and details of support resources as provided in the initial information will be repeated (see appendix 10 for paper version of debrief). The survey should take approximately 30 minutes to complete. Data from the online survey will be gathered electronically, and data from the hard copies will be inputted immediately into the electronic dataset. The hard copies of the questionnaire will be safely and immediately destroyed.

### **Proposed analysis**

A series of mediation analyses using Hayes PROCESS tool (Hayes, 2013) within SPSS will be used to examine whether coping style mediates the relationship between stigma and psychological outcomes.

**Practical issues**

For participants who would prefer to access the survey in paper format, an additional cost of postage will be incurred. This will be funded by the DClinPsy course at Lancaster University.

**Ethical concerns**

Participating in this study should not pose a significant risk to participants. However, reflecting on the issues raised within the research may be difficult for some people. Contact details will be provided of places that can be provide support should any issues arise. Additionally, it will be made clear prior to starting the study that they can stop at any time during the survey although, due to the anonymity of participation, their data cannot be removed after they have agreed to take part.

**Service User Involvement**

Two experts by experience have been consulted on the measures used within the survey and the design of the project. Changes were then made following feedback.

**Timescale**

**Ethics application:** 10<sup>th</sup> July 2019

**Recruitment** September 2019- January/Feb 2020

**Analysis and write up** February- April 2020

**Submission** May 2020

**Appendices**

Appendix 1: Participant information sheet. – paper version

Appendix 2: Participant consent form. – paper version

Appendix 3: Demographic and Clinical information – paper version.

Appendix 4: Stigma Scale for Chronic Illness (SSCI-24) (Rao et al., 2009) – paper version.

Appendix 5: Brief COPE (Carver, 1997) – paper version.

Appendix 6: Cervical Dystonia Impact Profile (CDIP-58) (Cano et al., 2004) – paper version.

Appendix 7: Warwick-Edinburgh Mental Well-being Scale (WEMWBS) (Tennant et al., 2007)- paper version.

Appendix 8: Depression Anxiety Stress Scale – 21 (DASS-21) (Henry & Crawford, 2005; Lovibond & Lovibond, 1995) – paper version.

Appendix 9: Functional Disability Questionnaire (FDQ) (Jahanshahi & Marsden, 1990) – paper version.

Appendix 10: Participant debrief – paper version

Appendix 11: link to online survey

Appendix 12: Advertisement for recruitment

Appendix 13: Video advertising research

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## Appendix 4-1 Approval letter



Applicant: Helen Gowling  
Supervisor: Fiona Eccles  
Department: Health Research  
FHMREC Reference: FHMREC18109

30 August 2019

Dear Helen

**Re: Stigma, coping styles and wellbeing in individuals with cervical dystonia**

Thank you for submitting your research ethics 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:- 01542 593987

Email:- [fhmresearchsupport@lancaster.ac.uk](mailto:fhmresearchsupport@lancaster.ac.uk)

Yours sincerely,

A handwritten signature in black ink that reads "R.E. Case".

Becky Case  
Research Ethics Officer, Secretary to FHMREC.

**Appendix 4-2 Amendment application****Faculty of Health and Medicine Research Ethics Committee (FHMREC)****Lancaster University Application for Amendment to  
Previously Approved Research**

1. Name of applicant:

Helen Gowling

[h.gowling@lancaster.ac.uk](mailto:h.gowling@lancaster.ac.uk)

2. E-mail address and phone number of applicant:  
3. Title of project:

**Stigma, coping styles and wellbeing in individuals with cervical dystonia**

4. FHMREC project reference number:

FHMREC18109

5. Date of original project approval as indicated on the official approval letter  
(month/year):

30/08/2019

6. Please outline the requested amendment(s)

Expansion to recruitment strategy to include:

Video to advertise the study on the websites of both Dystonia Ireland and the Dystonia Society.

For both charities to be able to be able to advertise through all channels available to them (i.e posters, newsletter, cascading to local groups)

Other dystonia relevant online organisations may also be approached to request the advert be shared through their channels.

**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**

7. Please explain your reason(s) for requesting the above amendment(s):

Following discussions with the Dystonia Society, they requested the video to be put on their website and suggested advertising in non-electronic ways as the people who access their service and may wish to participate in research do so in a variety of ways rather than just on social media. This also justifies expanding the recruitment to other dystonia relevant organisations as well as for the charities to advertise via other mediums besides electronically.

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 [Becky Case](#) There is no need to resubmit the Governance Checklist

**Applicant electronic signature:**

Date

Helen Gowling

07/10/2019

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):    Date application discussed

Fiona Eccles

08/10/2019

***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***

**Appendix 4-3 Amendment approval letter**

Applicant: Helen Gowling  
Supervisor: Fiona Eccles  
Department: Health Research  
FHMREC Reference: FHMREC19013

15 October 2019

Dear Helen

**Re: Stigma, coping styles and wellbeing in individuals with cervical 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 the amendment to 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:- 01542 593987

Email:- [fhmresearchsupport@lancaster.ac.uk](mailto:fhmresearchsupport@lancaster.ac.uk)

Yours sincerely,

A handwritten signature in black ink that reads "Becky Case".

Becky Case  
Research Ethics Officer, Secretary to FHMREC.

## Appendix 4-4 Online Survey

Online Survey Software | Qualtrics Survey Solutions

20/09/2020, 10:54

### Participant Information

#### Stigma, coping styles and well-being in individuals with cervical dystonia

Thank you for your interest in this research project. My name is Helen Gowling and I am conducting this research as a student in the Doctorate in Clinical Psychology programme and Lancaster University, Lancaster, United Kingdom

#### What is the study about?

I am interested in how you have experienced the attitudes of other people to cervical dystonia and any difficulties these may have created (stigma). I am also interested in whether these experiences have impacted on how you manage or cope with stressful situations. The aim is to understand the relationship between coping style, stigma and well-being.

#### Why have I been approached?

You have been approached because we need information from people who are over the age of 18 and have been diagnosed with neck dystonia/ cervical dystonia/ spasmodic torticollis.

#### Do I have to take part?

No. It is completely up to you to decide if you want to take part in the research. You can withdraw at any point before submitting the survey. Because your answers will be anonymous we will be unable to withdraw your responses after you have started to take part in the survey.

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

If you decide you would like to take part in this research you will be asked to complete a survey. This can be accessed online or on paper and will take approximately 30 minutes to complete. Paper copies can be made available on request (by phone: +44 7852 516812, by email: [h.gowling@lancaster.ac.uk](mailto:h.gowling@lancaster.ac.uk)). The survey will ask you questions about your condition, your feelings, the attitudes of others towards you and how you cope with stress. The online survey does not have to be

completed in one sitting, you are able to save your progress. You have 2 weeks from when you start to complete the survey. After this time the responses you have made will automatically be submitted and you will not be able to return to the survey.

**Will my data be identifiable?**

The data you provide will be completely anonymous. No one will have access to any personal information that identifies you. Lancaster University will store the electronic data from the survey for ten years. Information from paper copies will be inputted onto the electronic data source and the paper version will be immediately destroyed.

For further information about how Lancaster University processes personal data for research purposes and your data rights please visit our [web page](#).

**What will happen to the results?**

The results will be analysed and reported in a thesis which may be presented at conferences and/or be submitted for publication in an academic journal. A brief report of the findings will also be shared with the Dystonia Society and Dystonia Ireland. If you would like a copy of the report to be sent to you directly please contact Helen Gowling (by phone: +44 7852516812, by email: [h.gowling@lancaster.ac.uk](mailto:h.gowling@lancaster.ac.uk)).

Raw data will be kept confidential and only accessed by the research team. However, aggregated data may be shared with other genuine researchers if requested once the project is complete.

**Are there any risks?**

It is anticipated that there are no risks involved in participating in this research. However, if you experience any distress during or following participation, for example by answering difficult questions about your condition, you are encouraged to contact the resources provided at the end of this page.

**Are there any benefits to taking part?**

You may find taking part in the study interesting. The findings of the study may increase our understanding of stigma and inform ideas on how this can be addressed for people living with cervical dystonia.

**Who has reviewed the project.?**

This study has been reviewed and approved by the Faculty of Health and Medicine Research Ethics Committee at Lancaster University.

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

If you have any questions about the study, please contact the primary researcher Helen Gowling (y phone: +44 7852 516812, by email: [h.gowling@lancaster.ac.uk](mailto:h.gowling@lancaster.ac.uk)). The study will be supervised by Dr Fiona Eccles at Lancaster University and Dr Fiadhnaid O'Keeffe at St Vincent's University Hospital, Dublin.

**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:

Title: Professor Bill Sellwood Tel: (+441524) 593998

Email: [b.sellwood@lancaster.ac.uk](mailto:b.sellwood@lancaster.ac.uk)

Health Research

Lancaster University

Lancaster

LA1 4YG

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

Professor Roger Pickup Tel: +441524 593746

Associate Dean for Research Email: [r.pickup@lancaster.ac.uk](mailto:r.pickup@lancaster.ac.uk)

Faculty of Health and Medicine

Lancaster University

Lancaster

LA1 4YG

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

**Resources in the event of distress**

Should you feel distressed either as a result of taking part, or in the future, the following resources may be of assistance:

**The Dystonia Society (UK)**

<https://www.dystonia.org.uk/>

Tel: 020 7793 3651

Email: [support@dystonia.org.uk](mailto:support@dystonia.org.uk)

**Dystonia Ireland**

<http://www.dystonia.ie/>

Tel: 00 353 (01) 4922514

Email: [info@dystonia.ie](mailto:info@dystonia.ie)

**Your own GP**

If you experience distress as a result of taking part in this research we recommend that you seek support from your GP.

To download a copy of this information to keep please click on the link below:

[Participant information](#)

→

Powered by Qualtrics 

**Consent Form****Stigma, coping styles and well-being in individuals with cervical dystonia.**

In order to give consent to take part in the study please respond to the following statements by ticking the box to show you agree. If you have any questions or queries before signing the consent form please contact the principal investigator, Helen Gowling (by phone: +44 7852 516812, by email: h.gowling@lancaster.ac.uk).

I am over the age of 18

yes

I have received a diagnosis of cervical dystonia/ neck dystonia/ spasmodic torticollis

Yes

I confirm that I have read the information provided and fully understand what is expected of me within this study.

Yes

I understand that any responses/ information I give will remain anonymous

---

Yes

I understand that my participation is voluntary.

Yes

I understand that the researcher will discuss data with their supervisors as needed.

Yes

I understand that once I have started the survey I have 2 weeks to complete it.

Yes

I understand that my answers will automatically be submitted 2 weeks after I start the survey.

Yes

I understand that my answers cannot be withdrawn from the survey once I have started.

Yes

I consent to Lancaster University keeping the anonymised data for a period of 10 years

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after the study has finished.

Yes

I consent to take part in the study

Yes

How old are you? (Minimum 18 years)

What is your gender?

Male

Female

Other

What age were you when you first noticed symptoms of cervical dystonia?

At what age did you receive a diagnosis of cervical dystonia?

Are you taking prescribed medication to manage the symptoms of cervical dystonia? (if yes, please provide details)

Yes

No

Don't know

Have you received botulinum toxin injections to manage the symptoms of cervical dystonia?

Yes

No

Don't know

Are you receiving any other treatment to manage the symptoms of cervical dystonia?

Yes

No

Don't know

Do you currently have any other diagnosed physical or mental health conditions?

Yes

No

Don't know

What is your ethnic group? (you may choose more than one)

White English

White Welsh

White Scottish

White Northern Irish

White British

White Irish

White Irish Traveller

Any other White background

Mixed White and Black Caribbean

Mixed White and Black African

Mixed White and Asian

Any other Mixed/ Multiple ethnic background

Asian British

Asian Pakistani

Asian Bangladeshi

Asian Indian

Asian Chinese

Any other Asian background

Black British

Black African

Black Caribbean

Any other Black/ African/ Caribbean background

Arab

Other

Are you currently in paid employment?

Yes - Full time

Yes- Part time

No

Retired

What is your partnership status?

Single

Married/ Have a partner

Widowed

Other

What are your living arrangements?

Alone

With others (e.g. spouse, partner, children, friends, family)

Residential/ Nursing home

Other

Please read the following statements and indicate your level of agreement.

	Select your response				
	Never	Rarely	Sometimes	Often	Always
Because of my condition, I felt emotionally distant from other people	<input type="radio"/>				
Because of my condition, I felt left out of things.	<input type="radio"/>				
Because of my condition, I felt embarrassed in social situations.	<input type="radio"/>				
Because of my condition, I worried about other people's attitudes towards me.	<input type="radio"/>				
I was unhappy about how my condition affected my appearance.	<input type="radio"/>				
Because of my condition, it was hard for me to stay neat and clean.	<input type="radio"/>				
Because of my condition, I worried that I was a burden to others.	<input type="radio"/>				
I felt embarrassed about my condition.	<input type="radio"/>				
I felt embarrassed because of my physical limitations.	<input type="radio"/>				
I felt embarrassed about my speech.	<input type="radio"/>				
Because of my condition, I felt different from others.	<input type="radio"/>				
I tended to blame myself for the problem.	<input type="radio"/>				

I avoided making new friends to avoid telling others about my condition.	<input type="radio"/>				
Because of my condition, some people seemed uncomfortable with me.	<input type="radio"/>				
Because of my condition, some people avoided me.	<input type="radio"/>				
Because of my condition, people were unkind to me.	<input type="radio"/>				
Because of my condition people make fun of me.	<input type="radio"/>				
Because of my condition, people avoided looking at me.	<input type="radio"/>				
Because of my condition, strangers tended to stare at me.	<input type="radio"/>				
Because of my condition, I was treated unfairly by others.	<input type="radio"/>				
Because of my condition, people tended to ignore my good points.	<input type="radio"/>				
Some people acted as though it was my fault I have this condition.	<input type="radio"/>				
People with my condition lost their jobs when their employers found out.	<input type="radio"/>				
I lost my friends by telling them I have this condition.	<input type="radio"/>				

These questions deal with ways you've been coping with the stress related to living with cervical dystonia over the **past month**. There are many ways to try to deal with problems. These items ask what you've been doing to cope with this one. Obviously, different people deal with things in different ways, but I'm interested in how you've tried to deal with stress.

Each item says something about a particular way of coping. I want to know to what extent you've been doing what the item says. How much or how frequently. Don't answer on the basis of whether it seems to be working or not—just whether or not you're doing it. Try to rate each item separately in your mind from the others. Make your answers as true FOR YOU as you can.

	Select your response			
	I haven't been doing this at all	I've been doing this a little bit	I've been doing this a medium amount	I've been doing this a lot
I've been turning to work or other activities to take my mind off things.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I've been concentrating my efforts on doing something about the situation I'm in.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I've been saying to myself "this isn't real."	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I've been using alcohol or other drugs to make myself feel better.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I've been getting emotional support from others.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I've been giving up trying to deal with it.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I've been taking action to try to make the situation better.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

I've been refusing to believe that it has happened.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I've been saying things to let my unpleasant feelings escape.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I've been getting help and advice from other people.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I've been using alcohol or other drugs to help me get through it.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I've been trying to see it in a different light, to make it seem more positive.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I've been criticizing myself.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I've been trying to come up with a strategy about what to do.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I've been getting comfort and understanding from someone.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I've been giving up the attempt to cope.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I've been looking for something good in what is happening.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I've been making jokes about it.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I've been doing something to think about it less, such as going to movies, watching TV, reading, daydreaming, sleeping, or shopping.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I've been accepting the reality of the fact that it has happened.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I've been expressing my negative feelings.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I've been trying to find comfort in my religion or spiritual beliefs.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I've been trying to get advice or help from other people about what to do.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I've been learning to live with it.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I've been thinking hard about what steps to take.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I've been blaming myself for things				

that happened.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I've been praying or meditating	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I've been making fun of the situation.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

People with cervical dystonia can often be bothered by different problems. The following questions ask about problems you may have been bothered by **during the past 2 weeks**.

During the **past 2 weeks**, how much were you bothered by each of the following problems? (Please choose the option that best describes your situation.)

	Select your response				
	Not at all	A little	Moderately	Quite a bit	Extremely
a) Uncontrollable movements of your neck preventing your head from being straight?	<input type="radio"/>				
b) Twisting of the neck?	<input type="radio"/>				
c) Inability to control your head?	<input type="radio"/>				
d) Tension in your neck?	<input type="radio"/>				
e) Straining in your neck?	<input type="radio"/>				
f) Stiffness in your neck?	<input type="radio"/>				
g) Aching in your shoulders?	<input type="radio"/>				
h) Shoulder pain?	<input type="radio"/>				
i) Neck and shoulders being tired?	<input type="radio"/>				
j) Tightness in your neck?	<input type="radio"/>				
k) Tightness in your shoulders?	<input type="radio"/>				

During the **past 2 weeks**, has cervical dystonia limited your ability to carry out your usual daily activities?

	Select your response				
	Not at all	A little	Moderately	Quite a bit	Extremely
a) Limits in the type of work or other activities?	<input type="radio"/>				
b) Carrying heavy objects?	<input type="radio"/>				
c) Carrying light objects?	<input type="radio"/>				
d) Heavy household chores?	<input type="radio"/>				
e) Light household chores?	<input type="radio"/>				
f) Cleaning the house?	<input type="radio"/>				
g) Cooking?	<input type="radio"/>				
h) Getting tired when doing demanding physical activities?	<input type="radio"/>				
i) Getting tired when doing light physical activities?	<input type="radio"/>				

During the **past 2 weeks**, how much has your cervical dystonia:

	Select your response				
	Not at all	A little	Moderately	Quite a bit	Extremely
a) Limited your ability to walk?	<input type="radio"/>				
b) Limited your ability to climb up and down stairs?	<input type="radio"/>				
c) Limited how far you are able to walk?	<input type="radio"/>				
d) Increased the effort needed for you to walk?	<input type="radio"/>				
e) Slowed down your walking?	<input type="radio"/>				
f) Affected how smoothly you walk?	<input type="radio"/>				
g) Made you concentrate on your walking?	<input type="radio"/>				
h) Made you feel unsafe walking up and down stairs?	<input type="radio"/>				
i) Made you feel unsteady walking?	<input type="radio"/>				

During the **past 2 weeks**, how often did you:

	Select your response				
	None of the time	A little of the time	Some of the time	Most of the time	All of the time
a) Have trouble falling asleep because of the symptoms of your spasmodic torticollis?	<input type="radio"/>				
b) Have a restless sleep because of the symptoms of your spasmodic torticollis?	<input type="radio"/>				
a) Wake up because of the symptoms of your spasmodic torticollis?	<input type="radio"/>				
d) Not get the amount of sleep that you needed because of the symptoms of your spasmodic torticollis?	<input type="radio"/>				

During the **past 2 weeks**, has cervical dystonia limited your ability to carry out your usual social activities?

	Select your response				
	Not at all	A little	Moderately	Quite a bit	Extremely
a) Enjoyment of social situations?	<input type="radio"/>				
b) Socialising with friends or family?	<input type="radio"/>				

During the **past 2 weeks**, how often has cervical dystonia caused you to feel:

	Select your response				
	None of the time	A little of the time	Some of the time	Most of the time	All of the time
Angry?	<input type="radio"/>				
Annoyed?	<input type="radio"/>				
Irritated?	<input type="radio"/>				
Aggravated?	<input type="radio"/>				
Fed up?	<input type="radio"/>				
Frustrated?	<input type="radio"/>				
Stressed?	<input type="radio"/>				
Impatient?	<input type="radio"/>				
Upset?	<input type="radio"/>				
Worried?	<input type="radio"/>				
Anxious?	<input type="radio"/>				
Scared?	<input type="radio"/>				
Fearful?	<input type="radio"/>				
Depressed?	<input type="radio"/>				
Down?	<input type="radio"/>				
More self-conscious in social situations?	<input type="radio"/>				
Uneasy					

talking to strangers?	<input type="radio"/>				
Less relaxed in social situations?	<input type="radio"/>				
Embarrassed about eating in public (e.g. cafe, restaurant)?	<input type="radio"/>				
Embarrassed going out in public (e.g. cinema, theatre)?	<input type="radio"/>				
Everybody is staring at you?	<input type="radio"/>				
Lack of confidence?	<input type="radio"/>				
Lack of self-confidence?	<input type="radio"/>				

Below are some statements about feelings and thoughts. Please tick the box that best describes your experience of each over the past **2 weeks**.

	Select your response				
	Never	Rarely	Sometimes	Often	Always
I've been feeling optimistic about the future.	<input type="radio"/>				
I've been feeling useful.	<input type="radio"/>				
I've been feeling relaxed.	<input type="radio"/>				
I've been feeling interested in other people.	<input type="radio"/>				
I've had energy to spare.	<input type="radio"/>				
I've been dealing with problems well.	<input type="radio"/>				
I've been thinking clearly.	<input type="radio"/>				
I've been feeling good about myself.	<input type="radio"/>				
I've been feeling close to other people.	<input type="radio"/>				
I've been feeling confident.	<input type="radio"/>				
I've been able to make up my own mind about things.	<input type="radio"/>				
I've been feeling loved.	<input type="radio"/>				
I've been interested in new things.	<input type="radio"/>				
I've been feeling cheerful.	<input type="radio"/>				

Please read each statement and indicate how much the statement applied to you **over the past week**. There are no right or wrong answers. **Do not spend too much time on any statement.**

	Select your response			
	Did not apply to me at all	Applied to me to some degree, or some of the time	Applied to me to a considerable degree, or a good part of time	Applied to me very much, or most of the time
I found it hard to wind down.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I was aware of dryness of my mouth.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I couldn't seem to experience any positive feeling at all.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I experienced breathing difficulty (for example, excessively rapid breathing, breathlessness in the absence of physical exertion).	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I found it difficult to work up the initiative to do things.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I tended to over-react to situations.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I experienced trembling (for example, in the hands).	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I felt that I was using a lot of nervous energy.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I was worried about situations in which I might panic and make a fool of myself.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I felt that I had nothing to look forward to.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I found myself getting agitated.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

I found it difficult to relax.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I felt down-hearted and blue.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I was intolerant of anything that kept me from getting on with what I was doing.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I felt I was close to panic.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I was unable to become enthusiastic about anything.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I felt I wasn't worth much as a person.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I felt that I was rather touchy.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I was aware of the action of my heart in the absence of physical exertion (for example, sense of heart rate increase, heart missing a beat).	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I felt scared without any good reason.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I felt that life was meaningless.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

Below is a list of activities. Please indicate the extent to which your condition affects your engagement in, your performance or enjoyment of these activities **at the present time**.

	Select your response				
	Not applicable	Not at all affected	Mildly affected	Moderately affected	Severely affected
Dressing/undressing yourself	<input type="radio"/>				
Doing housework (vacuuming, washing, dusting, ironing etc.)	<input type="radio"/>				
Watching television	<input type="radio"/>				
Running	<input type="radio"/>				
Use of public transport	<input type="radio"/>				
Writing	<input type="radio"/>				
Having a face-to-face conversation	<input type="radio"/>				
Carrying objects	<input type="radio"/>				
Going to restaurants or pubs	<input type="radio"/>				
Brushing teeth	<input type="radio"/>				
Reading	<input type="radio"/>				
Walking	<input type="radio"/>				
Having sexual intercourse	<input type="radio"/>				
Driving a car	<input type="radio"/>				

Washing face	<input type="radio"/>				
Eating, using knife and fork	<input type="radio"/>				
Going to or giving dinner parties	<input type="radio"/>				
Typing	<input type="radio"/>				
Engagement in hobbies (knitting, sewing, carpentry, gardening, painting etc.)	<input type="radio"/>				
Crossing roads	<input type="radio"/>				
Shaving face if male, and putting make-up on, if female	<input type="radio"/>				
Drinking from a cup	<input type="radio"/>				
Riding a bicycle	<input type="radio"/>				
Going to the theatre/cinema/concerts	<input type="radio"/>				
Activities requiring visual/manual coordination such as pouring tea, using a screwdriver	<input type="radio"/>				
Engagement in sports (tennis, squash, jogging, swimming, golf, table tennis etc.)	<input type="radio"/>				
Walking up or down stairs	<input type="radio"/>				

Thank you for taking to the time to complete this survey.

It has been suggested that the way people cope with the stress of stigma can impact on their overall psychological well-being and quality of life. Therefore, this study aims to further examine this relationship and how it may apply to people living with cervical dystonia.

All the information collected from the surveys will be anonymous and there will be no way of identifying your responses in the data. If you have any questions about the study please don't hesitate to contact me on [h.gowling@lancaster.ac.uk](mailto:h.gowling@lancaster.ac.uk).

If you would like a copy of the summary report for this study you can request this by either email ([h.gowling@lancaster.ac.uk](mailto:h.gowling@lancaster.ac.uk)) or phone (+44 7852516812). A report will also be provided to The Dystonia Society and Dystonia Ireland. Please note this is a summary of all the data and I cannot provide reports based on individual responses to the survey.

Should you feel distressed either as a result of taking part, or in the future, the following resources may be of assistance:

**The Dystonia Society (UK)**

<https://www.dystonia.org.uk/>

Tel: 020 7793 3651

Email: [support@dystonia.org.uk](mailto:support@dystonia.org.uk)

**Dystonia Ireland**

<http://www.dystonia.ie/>

Tel: 00 353 (01) 4922514

Email: [info@dystonia.ie](mailto:info@dystonia.ie)

**Your own GP**

If you experience distress as a result of taking part in this research we recommend that you seek support from your GP.

**Appendix 4-5: Recruitment advert**

# Research Invitation

- Do you have a diagnosis of neck dystonia (cervical dystonia)?
- Are you over the age of 18?
- Could you spare 30 minutes to complete a survey about some of your experiences, thoughts and feelings?

My name is Helen Gowling and I am a doctoral student on the Clinical Psychology course at Lancaster University. This project aims to better understand the relationship between stigma, coping styles and well-being for people living with neck dystonia.

Further information and the survey can be found online by going to:

**<http://bit.ly/neckdystonia>**

Alternatively, paper copies are also available. If you would like to request one, or have any further questions, please get in touch:

☎ phone: +44 7852 516812

✉ email: [h.gowling@Lancaster.ac.uk](mailto:h.gowling@Lancaster.ac.uk)