Doctorate in Clinical Psychology | Lancaster University

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

March 2023

Doctoral Thesis

Psychosocial aspects of living with a visible neurological condition

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Prepared for: Journal of Health Psychology

Statement of total word count

	Word Count		
	Main text	Appendices (inc. tables, figures, references)	Total
Thesis Abstract	237		237
Literature Review	7,967	12,549	20,516
Research Paper	7,693	12,363	20,056
Critical Appraisal	3,688	1,467	5,155
Ethics Section	5,633	2,092	7,725
Total	25,218	28,471	53,689

Thesis abstract

This thesis examines the psychosocial aspects of experience for people living with visible neurological conditions.

Section one reports on a systematic literature review of qualitative studies exploring how individuals and families cope with Tourette's syndrome. A systematic search using keywords related to coping and Tourette's syndrome was conducted on four academic databases. A meta-ethnographic approach led to the construction of three themes: redefining the self and social identity; controlling the body; and challenging the narrative. The findings support a biopsychosocial approach to understanding the condition. This has clinical implications for the treatment of Tourette's syndrome and future research should seek to expand on this knowledge.

Section two reports on an empirical study exploring how people with neck dystonia navigate the social world. Ten participants were interviewed using a semi-structured, qualitative approach. Three themes were constructed from the data: dismissed by others for having an unfamiliar condition; negotiating a new social identity; and managing the stigma of a visible condition. The findings highlight the importance of social identity and the impact of stigma on people with visible health conditions. Further research should seek to explore the nature of distress arising from these psychosocial difficulties with the aim of tailoring clinical interventions for people with neck dystonia.

Section three includes a critical appraisal with reflections on the process of conducting this project. Consideration is also given to the role of psychology in addressing systematic societal concerns such as stigma.

Declaration

This thesis was undertaken between August 2021 and March 2023 as part requirement of the Lancaster University Doctorate in Clinical Psychology. The work documented here is my own except where due reference has been made in the text. This thesis has not been submitted for an award of a higher degree elsewhere.

Signature:

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Date: 10 March 2023

Acknowledgements

I would like to thank the participants who took part in the empirical study for being so generous and open in sharing their experiences with me. I would like to thank Dystonia UK and the various individuals who shared their expert advice and gave practical help.

I would also like to thank Dr Fiona Eccles and Dr Fiadhnait O'Keefe for the supervision they provided throughout the thesis process, and for all the feedback, encouragement and support.

Finally, I want to thank my husband and my children for all their understanding and patience throughout the whole of my doctorate and for making this possible for me.

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Section One: Literature Review

Coping with Tourette's syndrome: A meta-ethnography of individual and family perspectives

Word Count – 7,967
(excluding title page, references, figures, tables and appendix)

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Abstract

Tourette's syndrome is a neurological condition characterised by motor and phonic tics. This systematic review and meta-ethnography aimed to examine how children, adults and families cope with Tourette's syndrome. A systematic search of four databases was completed in October 2022. Analysis of 16 papers using Noblit and Hare's (1988) meta-ethnographic approach led to the construction of three themes: redefining the self and social identity, controlling the visible presentation of Tourette's syndrome, and challenging the narrative. Clinical implications of the findings are considered, including integrating complementary psychotherapy approaches into behavioural treatments, and future avenues for research are proposed.

Keywords: children and young people, adults, parents, families, Tourette's syndrome, tics, coping, systematic review, meta-ethnography

Introduction

Tourette's syndrome (TS) is a childhood-onset, neurological condition, classified by several motor and at least one phonic tic, lasting for longer than one year (DSM-5, 2013). A tic is a repetitive, non-rhythmic motor movement or vocalisation involving discrete muscle groups (DSM-5, 2013). TS symptoms typically begin as simple motor tics, such as eye blinking or facial grimaces, often becoming more complex and vocal with time (Leckman et al., 1998). The onset of tics typically occurs between the ages of four and six years old and reaches peak severity between the ages of ten and 12 years (Leckman et al., 1998). Tic severity generally declines during adolescence (Leckman et al., 2006). Roughly one half to two thirds of children have significant improvement of TS by adulthood (Bloch & Leckman, 2009). However, the remaining adult cases are thought to represent severe cases with persistent and complex tics (Cheung et al., 2007).

The prevalence of TS is estimated to be around 0.3 – 0.9 per cent for school-age children (Scharf et al., 2015), and 118 cases per one million adults (Levine et al. 2019). Prevalence is thought to be four times higher in males than females during childhood (Yang et al., 2016). However, the risk ratio of male to female reduces during adulthood to 2.33 (Levine et al., 2019). This could suggest girls have more persistent symptoms than boys and/or that female cases are not diagnosed so readily during childhood (Levine et al., 2019). One explanation for this is that females are more likely to perform 'internalised' tics than males which makes diagnosis more difficult (Garcia-Delgar et al., 2020).

The exact aetiology of TS is still unknown, yet is likely to feature both genetic and environmental factors, such as post-infectious autoimmunity and prenatal difficulties (Pashou,

2013). The type, frequency and intensity of tics are influenced by internal and external factors affecting the individual, such as emotional states and behavioural reinforcers (Gagne, 2019). Tics can be exacerbated by pain, tiredness, excitability, talking about tics and witnessing others' tics (Caurin et al., 2014). Tics can often be alleviated if the individual engages in enjoyable physical or mental activity that requires concentration (Caurin et al., 2014). Individuals are sometimes able to suppress tics, for example, during clinical appointments and in school (Conelea & Woods, 2008). However, this can cause increased pain, and in the aftermath of a period of tic suppression, can lead to 'tic attacks', involving the repetition of tics for hours at a time (Collicott et al., 2013).

Treatments for TS include psychoeducation, behavioural therapy, medication and in rare cases neurosurgery (Robertson et al., 2017). The first step in the treatment pathway often involves providing reassurance to the individual and their family, alongside relevant and accurate information about the nature and prognosis of TS (Cavanna & Seri, 2013). Evidence also supports the early application of habit reversal therapy as part of a wider comprehensive behavioural intervention for tics, for both children (Piacentini et al., 2010) and adults (Wilhelm et al., 2012). For individuals who are experiencing considerable functional impairment due to TS, such as pain and sustained social and emotional difficulties, alpha-adrenergic agonists and anti-psychotics can be considered alongside behavioural interventions (Cavanna et al., 2013). However, TS is difficult to treat with medication and the potential side effects of some treatments can affect quality of life and social interaction (Hartmann et al., 2016).

Treating TS is complex, and this is in large part due to the common co-morbidities (Cavanna, 2018). Indeed, Cavanna (2018) emphasises how TS is a cluster of multiple phenotypes rather than a unitary condition as originally believed; and that this has led to a

"paradigm shift" in the understanding of TS. Over half of individuals with TS also have attention-deficit hyperactivity disorder (ADHD; Khalifa, 2006) and roughly one-third to one-half of children with TS will experience obsessive-compulsive disorder (OCD) throughout their lifetime (Bloch et al., 2006). Approximately 40 per cent of children with TS will also experience depression or anxiety (Gorman et al., 2010; Bloch et al., 2006). The combination of TS and co-occurring conditions can cause difficulties in concentration and academic learning for adolescents (Lee et al., 2016; Wadman et al., 2016). Adults with TS have been reported to commonly experience a range of psychological difficulties, including severe mood swings, problems with concentrating and sleep problems (Altman et al., 2009). A population cohort study in Sweden found people with TS are at an increased risk of suicide when compared with a control group, and that persistence in tics beyond young adulthood and previous suicide attempts were the strongest predictors of death by suicide (del la Cruz et al., 2017).

Psychological factors have been shown to be among the most important determinants of overall quality of life in children with TS (Cavanna et al., 2013) and in adulthood (Cavanna et al., 2008). Whether psychological difficulties are a component of TS or a consequence of living with tics is likely to be multifactorial (Robertson, 2006). For example, tics by their nature involve hyperactivity, not only the expression of tics but also the effort to actively suppress them, and this would likely have an impact on concentration (Cavanna et al., 2009). Studies have found that functional impairment is positively correlated with both tic severity and co-occurring conditions in children and adults (Gorman et al 2010; Eapen et al., 2016; Lin et al., 2007). However, some studies have found these co-occurring conditions cause more difficulties for individuals with TS than the tics themselves (Bernard et al., 2009).

Many individuals with TS experience a sensory phenomenon, known as a premonitory urge, that occurs immediately prior to a tic (Leckman et al., 1993). This is often described as feeling like an itch or a need to sneeze (Leckman et al., 1993). Awareness of premonitory urges increases with age and is present in as many as 90% of adolescents with TS (Woods et al., 2005). Often the urge is only relieved if the tic is actioned "just right" (Chowdhury & Murphy, 2016, p.21), leading to repeated tics until a feeling of satisfaction. However, such repetition can be exhausting and painful. Findings from the Tourette Syndrome Impact Survey showed the majority of children and adults had at least one tic that caused pain (Conelea et al., 2013; Conelea et al., 2011).

In addition to the biological and psychological features of TS, there are also social factors which impact on people living with the condition. For instance, TS is a largely misunderstood condition in society, often being inaccurately associated with people who repeatedly swear or say inappropriate phrases (Fat et al, 2012; Calder- Sprackman et al 2014). This is despite the prevalence of coprolalia (the uncontrollable utterance of obscenities and profanities) in people with TS is estimated to be 8.5% (Freeman et al., 2009). Lack of awareness and misinterpretation can lead to stigmatised attitudes towards people with TS (Malli et al., 2016). Many children with TS also face teasing and bullying, not only from peers but also from adults, including teachers, who do not understand the condition (Dempsey et al., 2018). A survey of 109 adults with TS found that respondents experienced discrimination in multiple life domains, including work and education (Malli & Forrester-Jones, 2022). Such discrimination included not being considered for job interviews and being denied access to training and reasonable adjustments (Malli & Forrester-Jones, 2022).

TS is something that affects the whole family, including increased risk of anxiety and depression for parents of children with TS (Cooper, 2003; Shoeder, 2007; Wilkinson, 2008; Woods et al., 2005). The stress felt by parents can negatively impact on parent-child relationships (Lee et al., 2007). Consequently, both families and individuals must adopt coping strategies for living with TS. Given that the stressors faced by children, adults, and families are multifaceted, the concept of coping with TS must be suitably broad to encompass all aspects. One such broad definition is that of Lazarus and Folkman (1984), who described coping as: 'constantly changing cognitive and behavioural effort to manage specific external and/or internal demands that are appraised as taxing or exceeding the resources of the person' (p. 141).

Lazarus and Folkman (1984) also make the distinction between problem-focused and emotion-focused coping. The former involves efforts to address the problem at hand, whereas the latter involves efforts to manage the emotional distress associated with the problem (Lazarus & Folkman, 1984). Coping often involves both problem and emotion-focused aspects, along with differing levels of control, as described by the control-based model of coping (Connor-Smith et al., 2000; Walker et al., 1997). This model distinguishes between primary control or active coping (efforts to act on the source of stress or associated emotions), secondary control or accommodative coping (efforts to adapt to the source of stress), and disengagement or passive coping (efforts to avoid or deny the stressor) (Connor-Smith et al., 2000; Walker et al., 1997). This review will take a broad approach by including all experiences of participants that show purposeful efforts to respond to TS-related stress, in addition to noting the level of control used. An inclusive approach to coping strategies will be taken, including, for example, cognitive strategies such as avoidance, humour and denial, strategies to control symptoms, and social strategies such as seeking out support, disclosure and self-advocacy

(Audulv et al., 2016). Similarly, the outcomes of these strategies will remain broad to encompass the widest range of experience. Outcomes could include acceptance and positive self-image, process skills such as problem solving, quality of life, control of symptoms and social participation (Audulv et al., 2016).

The effectiveness of these strategies depends on the match between characteristics of the stressor, especially perceived controllability, and the individual's coping responses (Compas et al., 2012). In quantitative studies, secondary control coping has been found to lead to successful adaptation to chronic illness in children and adolescents, whereas disengagement coping is associated with poorer adjustment (Compas et al., 2012). In terms of coping with TS, a cross-sectional survey of Canadian adults with TS found that family support was the most important coping strategy to respondents when they were younger than 18, whereas personal acceptance was the most important coping strategy over the age of 18 (Altman et al, 2009). However, quantitative evidence can only show the relative weight given to different coping strategies, and does not provide understanding of coping in a broader context.

A meta-synthesis exploring the lived experience of children and adults with tic disorders identified one theme that related to coping (Smith et al., 2015). The review found that adaptive coping strategies focused largely on tic management, including techniques to hide or disguise tics in response to the stigma that participants experienced (Smith et al., 2015). However, the review did not explore the theme of coping in depth. The aim of the present paper is to build on this meta-synthesis by focusing specifically on coping with TS and by including the perspectives of family members. Given the dynamically evolving understanding of TS, a meta-synthesis will bring together findings from the qualitative literature to explore the coping strategies of participants and to identify new interpretations of the data. Deeper

understanding of how individuals and their families cope can contribute to the evidence base for clinical interventions and can generate new research questions to advance the field. The purpose of the present review was to answer the following research question: How do people with Tourette's Syndrome, and their families, cope with the condition?

Method

A meta-ethnography was chosen as it is a well-developed model for both synthesising qualitative data and extending conceptual knowledge (Britten et al., 2002). The method provides a systematic approach which maintains the interpretive properties and contextual factors of the original papers (Dixon-Woods et al, 2005). This meta-ethnography of coping with TS followed the procedure of Noblit and Hare's (1988) seven-step approach. These steps are as follows: getting started, deciding what is relevant to the initial interest, reading the studies, determining how the studies are related, translating the studies into one another, synthesising translations and expressing the synthesis.

Search strategy

To identify appropriate studies, four electronic databases were systematically searched: PsycINFO, MEDLINE Complete, CINAHL and SocIndex. These were chosen with the aim of covering psychological, medical and sociological research. The searches were conducted in October 2022. Google Scholar was searched, and the references of key papers were also hand searched for additional studies (Sayers, 2007). To ensure no papers were missed, search terms focused on the two main concepts of TS and coping, using a range of alternative terms. Free

text searches were used in the title and abstract field of all databases, coupled with the relevant subject headings. Free text search terms for TS consisted of:

*Tourette** OR *tics* OR *"tic disorder"*

The free text search terms used for coping were:

Coping OR *adjustment* OR *wellbeing* OR *experience* OR *"living with"*

These were searched for within the title and abstract fields of all four databases. This was combined with the databases' own subject headings relevant to TS and coping using the Boolean operator "*OR"*. The searches relating to TS and to coping were then combined using the term "*AND"*. Limiters were used within the databases to exclude papers that were not written in English and were not peer reviewed. No limits were set on the date of publication. Table 1 -A shows the full list of search terms.

<Table 1 – A about here>

Studies had to meet the following inclusion criteria:

1. Written in English.
2. Published in a peer-reviewed journal.
3. Used a qualitative research design. Mixed methods research papers were included if it was possible to clearly separate the qualitative results from the quantitative ones. Studies were considered to be qualitative if participants were asked open-ended or semi open-ended questions (Dixon-Woods et al., 2005).
4. Included participants with Tourette's syndrome (children, adolescents and adults) or the family members of people with Tourette's syndrome.
5. Analysis derived from experience of Tourette's syndrome given from first person accounts.

6. Papers had at least one major theme (or concept) focused on coping with TS.

The following types of papers were excluded:

1. Papers which focused on the experience of diagnosis only.
2. Papers which focused exclusively on the experience of or evaluation of having treatment.
3. Papers which focused on the evaluation of services.
4. Qualitative papers which were not explicit about the method used to analyse the data.

The search strategy identified 1,613 studies. These were exported to Endnote (Clarivate Analytics, n.d.), which identified 889 duplicates. Title and abstract scan meant a further 690 papers were excluded due to not meeting the inclusion criteria or meeting the exclusion criteria. Full text copies of 34 papers were read and a further 18 were excluded, meaning 16 studies were selected for inclusion. The reference lists of included papers were also searched for any additional relevant papers. This process of screening is summarised in a PRISMA diagram (Moher et al., 2009) (Figure 1 - A).

<Figure 1 – A about here>

Characteristics of selected studies

Of the 16 papers included, 12 looked at individual perspectives and four focused on parent perspectives. Of the 12 individual papers, six focused on adults and six focused on children and adolescents. A total of 65 young people, 80 adults, and 123 parents and carers of young people with TS were included as participants. A range of cultural contexts were

included, with participants from the UK, USA, Canada, Australia, New Zealand and Taiwan. The studies covered a breadth of topics relating to the psychosocial experiences of living with TS and used a variety of qualitative approaches to data analysis. Details of included papers are summarised in Table 1 - B.

<Table 1 - B about here>

Quality appraisal

A Critical Appraisal Skills Programme (CASP) checklist was used to assess the quality of included studies (CASP, 2018). CASP was chosen as it creates a systematic process through which the strengths and weaknesses of each study can be identified (Scott & Grant, 2018). The checklist consists of two screening questions, then a further seven detailed questions and one open-ended question. These latter eight questions were scored using the three-point rating system of Duggleby et al. (2010). A weak score was given one point, a moderate score two points, and a strong score three points, giving a maximum of 24 points. This resulted in scores of between 15 and 22 (mean = 18.6). All papers were deemed to be of reasonable quality. Full results of the quality appraisal can be found in Table 1 - C. The aim of the meta-synthesis was to provide a comprehensive review of perspectives of coping, therefore studies with low quality scores were not excluded (Sandelowski & Barroso, 2007). Instead, the quality ratings were used to decide on an 'index study' (Atkins et al., 2008). This was the study with the highest score, and the first study from which codes were translated into other studies as a way of positively shaping the research.

<Table 1 - C about here>

Synthesising the selected studies

Following the selection and appraisal of the 16 studies, the next step was to carefully read each paper, extracting methodological details and contextual information. Following this, each paper was re-read so that major concepts and themes could be identified. These were noted down along with relevant quotes from participants and the authors' interpretations of these experiences. This information was recorded in a table in order to determine how the papers were related to each other. The aim was to keep the wording as close to the original authors' language in order to retain the meaning. The next stage of the synthesis involved developing second order interpretations. These interpretations were then synthesised leading to the final three themes. This was achieved by taking the findings from the index paper and comparing them to the next paper in the order of quality and conceptual richness. The resulting synthesis was then compared to the third study, and so on. Noblit & Hare (1988) refer to this as 'reciprocal translation'. The continual process of comparison of concepts enabled the development of the themes (Schutz, 1962). Table 1-D demonstrates the process of theme identification. All the resulting themes were present across several papers, with no themes depending solely on data from papers with a low CASP score (see Appendix 1- A for a summary of the synthesised themes). There were no papers that refuted the findings of another study. A line of argument synthesis was then conducted to show how the themes were related.

<Table 1 - D about here>

There is a risk of interpretation bias when conducting a meta-synthesis, given that the notion of total objectivity is unachievable in qualitative research (Tufford & Newman, 2012). It is important to acknowledge the researcher does not have any personal experience of TS and therefore her perceptions were shaped by cultural and social beliefs and biases related to the condition. However, any risk of bias was minimised by the researcher consulting with two supervisors throughout the synthesis stage in order to identify and explore preconceptions (Morse et al., 2002).

Results

In synthesising the 16 papers, three main themes relating to how individuals and parents cope with TS were constructed: (1) redefining the self and social identity (2) controlling the visible presentation of TS and (3) challenging the narrative.

Redefining the self and social identity

This theme relates to how people cope with TS in relation to threats concerning their self-identity and their social identity. TS, with its visible symptoms, marks people out as 'different' from others, and this has implications for identity management. For individuals, accepting TS as either part of the self or separate to the self can reduce the negative emotional impact of TS. Malli et al. (2019) identified two divergent groups of adult participants; the first had accepted TS as part of their identity, whereas the second group could not integrate the condition into their identity and instead resisted association with TS. For example, one participant in the first group noted:

"Because Tourette's is me and I've obviously come to terms with it and, you know, I embrace it…and, you know, I've learned to accept that that's just me." (Participant 9: Malli et al., 2019, p. 827)

A similar experience of integrating TS into self-identity was reported in adolescence (Cutler et al., 2009; Lee et al., 2019). The story of identity integration was associated with a narrative of personal growth and the outcome was one of positive coping (Smith et al., 2016; Malli et al., 2019). This process of learning to accept TS as part of self-identity during adolescence was aided by supportive friends and family, demonstrating the importance of this period of development (Lee et al., 2016; 2019; Wadman et al., 2013). The support from friends and family gave adolescents with TS the self-confidence (Lee et al., 2019) and courage (Lee et al., 2016) to see themselves in a new and positive light. However, there were also negative aspects of coping associated with integrating TS into self-identity, as TS was also described as having overtaken or diminished the individual's previous identity (Cutler et al., 2009; O'Connor et al., 2009).

When TS was perceived as being external to the self, as evidenced by participants' using externalising language such as "the tics", it evoked a negative, irritating presence: *"... annoying like a wasp is annoying, it never goes away ..."* (Participant 3: Smith et al., 2016, p1792). One participant described TS as an independent entity attached to her 'normal' self:

"I think Tourettes is like another person isn't it – it's like you've got me on my normal side and you've got to take like Tourettes with I". (Deborah: Wadman et al., 2013, p. 882)

Perceiving TS to be separate to themselves also had positive and negative consequences for coping. For instance, one participant aspired to learn from an actor who had successfully externalised their TS and come to see TS as their friend (Lee et al.,2016). On the other hand, there was concern that others could not see past the tics to see the individual for who they really are (O'Connor et al., 2009).

The concept of acceptance and integration into identity was also an important aspect of coping for parents of young people with TS. Accepting TS led to feelings of loss for some parents as they mourned the idea of an "*ideal child*" (O'Hare, 2016, p. 52). Although this may have led to helpful emotional processing, it also led to feelings of guilt, as parents acknowledged that TS was not life threatening (O'Hare, 2006, p. 52). Parents highlighted how important validation was, given the feelings of guilt involved. Validation could be expressed by others as acceptance of the diagnosis, that their child had a neurological condition, and through acknowledging how difficult it was to live with TS (Travis & Juarez-Paz, 2020). One mother noted how validating it was to hear from others that she was "*doing a great job*" and that TS "*isn't anything that [she] created*" (Travis & Juarez-Paz, 2020, p. 1485).

Individuals and parents also had to decide whether to join a TS community or not, thus acknowledging a TS social identity. Resisting identification with a TS group can also avoid having to identify with a stigmatised group (Malli et al., 2019). As an author reflecting on his own TS noted:

"Not acknowledging it [TS] and refusing to go to these support groups helped me think that I was an individual who was in control of what I wanted to do." (Congdon, 2014, p.15).

On the other hand, joining support groups, can lead to increased acceptance and validation from others "*who get it*" (O'Hare et al., 2017, p.53).

Controlling the visible presentation of TS

A major component of coping involved controlling the visible presentation of tics. This was regardless of whether the perspective was an individual or a parent one. Such control could be exerted internally, for example, by suppressing the tics (subtheme 1), or externally, for example, by removing the self or family member away from other people (subtheme 2).

Internal control

The need to conceal tics was driven by a desire to fit in and avoid negative attention (Cutler et al., 2009). Strategies to control the symptoms of TS such as ignoring, supressing or disguising tics were highly valued by individuals (Smith et al., 2016). This was even though supressing tics could cause pain and exhaustion (Lee et al., 2016). Perhaps due to the difficulty involved, such mastery over tics was a source of pride (Wadman et al., 2013). As one participant explained:

"It takes a lot of power [controlling tics] yeah, I'm not sure a lot of people can do that'. (Kurt: Wadman et al., 2013, p. 882).

Less direct ways of exerting control over tics included avoiding the internal physical and mental states that trigger tics, for example, stress and fatigue (Lee et al., 2016). On the other hand, some participants were able to coexist with their tics and learnt to resist the pressure to

hide them. For some, this came with age, and as they grew older, they gained more confidence and stopped caring what other people thought (Lee et al., 2016).

External control

Parental perspectives suggested a similar need to manage the visible presentation of tics. However, as parents themselves could not suppress the tic, they could still exert control over the visibility of the tics, for example, by avoiding social situations where other people would see their child's tics. One participant reported that they chose to go on holiday to quiet places with fewer people around (Ludlow et al., 2018). For some parents, it was not the visibility of the tics which required control, but the physical nature of the tics themselves, as one parent described:

"We have tea earlier than we would normally because … if we had tea later and her tics are really bad and she couldn't physically eat." (Helen: Ludlow et al., 2018, p.1794).

The constant requirement to conceal their children's tics and to manage the physical nature of tics was referred to as a "*daily struggle*" and the "*new normal*" (Travis & Juarez-Paz, 2020). In order to ease the struggle, home became a safe place, where individuals with tics did not feel required to regulate their tics in front of others (Malli & Forrester-Jones, 2021) and parents did not have to manage and monitor the reactions of strangers (O'Hare et al., 2017). As one parent explained:

"I suppose with our family (nuclear) unit, we are very strong with each other, we rely on each other a lot which I think helps in lots of ways. We said, 'honey you just tic as much as you want

and as often as you want and as loud as you want because we don't care' (mum, dad and siblings)." O'Hare et al., 2017, p. 53.

Spending more time at home to reduce the stress of being in public led to some families becoming more isolated (O'Hare et al., 2017). Although isolation could have potentially negative effects on wellbeing, this could also be seen as a protective strategy to avoid social rejection (Malli et al., 2019). As one participant said:

"…the tics really made me less likely to reach out socially. Well, to socialise at all or to seek out friends, 'cause I was worried about being the joke." (Participant 3: Malli et al., 2010, p. 834)

For parents, adaptations to the school environment were a major feature of helping their children to cope with the visibility and physical nature of the tics. Such adaptations were practical, for example, access to a private room to give school children space to tic on their own (Ludlow et al., 2018; Pine et al., 2022). Other adaptations included making sensory toys available, having extra time for exams or being able to take exams in a private room (Pine et al., 2022). Parents also stressed that more emotional support for children from teachers was necessary, so a relationship of collaboration and understanding could emerge (Pine et al., 2022). One parent noted how successful their school had been in being able to balance providing extra care without making the child appear different in front of peers (Ludlow et al., 2018).

Challenging the narrative

This theme identifies how one of the main difficulties with having TS or having a family member with TS was being misunderstood or dismissed by others. Misunderstandings about the nature of tics, and trivialisation of the impact of TS seemed a common experience for people, especially regarding the prominence of coprolalia, as one participant explained:

*"It's like Tourette's is only the f*** word, it's not the pain, not having to open your brain to make the pain go away. Just the swearing."* (Participant 5: Malli & Forrester-Jones, 2021, p. 13)

Participants discussed the importance of having to explain the involuntary nature of tics to others (Edwards et al., 2017) and some wanted others to understand the significant impact that tics had on them (Wadman et al., 2013). For one individual with TS, the need to educate people about the condition became all consuming, so much so it was difficult to be themselves in public:

"I have become pretty damn good at explaining what Tourette's is. But it's draining and it's like exhausting...I always have to be the 'Tourette's girl'" (Participant 20; Malli & Forrester-Jones, 2021, p. 17)

There was also a desire to explain the medical causes of tics (Cutler et al., 2009). This may have been because it felt less stigmatising than letting people conclude that tics have a psychological cause (Buckser, 2008). Such a need to communicate this to others was complicated by the competing need of having to control tics by suppressing them. This could

give the impression to others that the tics were controllable and therefore individuals were 'responsible' for their tics (Buckser, 2008).

Parents also struggled with a lack of understanding from others, which led to feelings of being belittled and disbelieved (Travis & Juarez-Paz, 2020). They expressed the need to educate others about the nature of Tourette's syndrome. This first meant taking time to become experts on the condition themselves, as "*understanding makes lives easier*" (Travis & Juarez-Paz, 2020, p. 1,484). Parents wanted others to know that Tourette's syndrome meant "*more than swearing*" (Pine et al., 2022, p. 6) and that it had a neurological cause:

"I know it's probably funny to watch on the TV when they're shouting out, you know, swear words … I think some people, you know, take the mick but … it's so tiring and when you see your son in that much pain because of it, erm, and he's just worn out … it's making people understand that it's not that funny." (Rose; Ludlow et al., 2018, p. 1,795)

Once equipped with expert knowledge about TS, parents discussed how they would use this to advocate for changes that would help their children. For example, one parent recommended teachers should:

"Increase . . . understanding of what an individual goes through' and 'teach kids that it's ok to have Tourette's." (Pine et al., 2022, p. 6)

Line of argument synthesis

As is evident from the themes identified, the focus of coping in this synthesis is coping with the stigma of having a visible condition, rather than coping with the physical problems associated with TS, or with associated neurodevelopmental conditions. The most prevalent theme across all the papers was the need to conceal the visible nature of tics from others. This was the case for young people and adults with TS and parents. However, a supportive social environment, which included adaptations at school, could lead some to feel pride that they were able to exert control over their tics. Parents could support this by helping to make home a safe space for their children to tic freely. A feeling of pride and supportive friends and family allowed individuals to assimilate TS into their identities. However, without a supportive social environment, having to conceal tics could lead to social withdrawal and isolation. This was more likely to lead to difficulties with accepting TS as part of individual identity. This line of synthesis argument illustrates how the themes are related (see Figure 1 - B).

<Figure 1 – B about here>

Discussion

The aim of this synthesis was to explore how individuals and families cope with TS. The results show how coping with TS was focused on coping with the visible presentation of tics, the associated stigma and the impact this has on self-identity. People with TS and their families belong to a society that recognises tics as differing from a social norm, and are therefore coping with having a "spoiled identity" (Goffman, 1963). Coping also relates to educating others about TS to reduce misrepresentations of the condition and in the case of

parents, to fight for support for their children at school. Interestingly all the papers included in the synthesis emphasised these psychosocial aspects of coping rather than coping with physical problems such as pain or fatigue. The line of argument synthesis suggests a new way of connecting stigma management strategies and identity development in individuals with TS with social support from families and wider social narratives.

The first theme highlighted how individuals with TS cope with the condition by either integrating or externalising it in relation to their own identity. In terms of coping theory, this renegotiating of identity would appear to be an emotion-focused approach, in that individuals are seeking internal strategies to adapt to the distress caused by the condition (Lazarus & Folkman, 1984). Acceptance from others allowed individuals to incorporate the TS identity which enabled positive self-evaluation, and even personal growth. Evident in the participants' accounts was a narrative that told of how they had overcome the difficulties of TS to become a better person. The construction of such narratives are important mechanisms to help individuals understand a health condition in terms of their past experiences and reaffirm a sense of self with purpose and meaning (Williams, 1984)

This could also relate to the Social Identity Model of Identity Change (SIMIC; Jetten et al., 2009), which stipulates that life transitions, such as the onset of health conditions, which lead to identity changes, can be positively adapted to when people are able to maintain pre-existing social group memberships or to develop new ones. Individuals reported that it was the support of family, friends, partners and teachers, that allowed them to renegotiate their identities and accept the condition as part of themselves (Cutler et al., 2009; Lee et al., 2016; 2018; Malli et al., 2019; Wadman et al., 2013).

The SIMIC could also be relevant given that many of the participants in the review were also in a transitional time from adolescence to adulthood (Kroger, 2006), meaning that the ability to remain part of friendship groups throughout the TS experience is particularly important. In addition to the benefits that social support can provide in terms of identity and acceptance (Haslam et al., 2005; Jetten et al., 2014), social support has also been found to moderate the effects of stigma on people with mental health conditions (Mueller et al., 2006; Verhaeghe et al., 2008). In the present review, positive social support allowed people to feel proud that they could control their tics, provided safe spaces to tic, and supported the acceptance of TS. Having to cope with stigma by concealing tics, but without social support, seemed to have a more negative impact on individual's sense of self and led to negative consequences such as social isolation (Lee et al., 2016; 2018).

The concept of control was the key feature of the second theme which highlighted how participants attempted to control the visibility of their tics, or in the case of parents, attempted to control their child's tics so that they were less visible or disruptive to daily activities. Ability to control the tics gave participants a sense of pride, which could relate to the theory of the 'locus of control' (Rotter, 1966). This stipulates that control over actions and events can be attributed to internal factors, such as an individual's characteristics or behaviour, or external factors, such as chance or the actions of others (Rotter, 1966). A higher belief in internal control has been associated with lower levels of depression in people with other neurological conditions (Jahanshahi, 1991). Verity et al (2020) found that perceived control, that is, the level of control felt by an individual over their health condition, in this case, Parkinson's disease, mediated the relationship between stigma and health related quality of life, depression and positive affect. The development of an internal locus of control in children could be related to parenting style. For example, Cohen et al. (2008) used a regression analysis with 65 children

with TS and found an internal locus of control, which is associated with an autonomy-granting parenting style, acted as a protective factor against anxiety and depression.

The third theme, which involved challenging the wider social narratives about TS and other people's misunderstandings, could be conceptualised as problem-focused (Lazarus & Folkman, 1984) and active coping (Connor-Smith et al., 2000; Walker et al., 1997). This also reflects the dilemma identified by Buckser (2006) who described TS as a condition which exists outside the standard classifications of how people in Western countries understand illness. For instance, people with TS experience the simultaneous presence of both voluntary and involuntary features of tics, as the urge to tic is beyond conscious control, the decision to release the tic is not (Cavanna & Nanni, 2013). This means it is difficult for people affected by TS to explain the condition in ways that society can understand (Buckser, 2006). However, if a condition cannot be suitably understood and explained to people then it becomes more likely to become stigmatised (Albrecht et al., 1982).

The findings suggest that the psychosocial aspects of coping with TS are key, specifically, how social environments act as both a stressor and a coping resource. The line of argument synthesis applies the SIMIC (Jetten et al., 2009) to TS, a visible neurological condition which is subject to the stigmatising views of others and the misrepresentations of societal narratives. However, as TS is a childhood onset condition, the extent to which the condition impacts on identity change must be understood within the context of a developmentally significant period (Erikson, 1968). The biographical disruption account describes how the onset of health conditions represents a re-evaluation of self-hood including perceptions of self, expectations and relationships (Bury, 1992). Alongside this, Charmaz (1983) describes how individuals with chronic illnesses experience a loss of self as daily

activities and social relationships which would normally reinforce self-concept are diminished. However, these theories are largely based in adult work and presume that a "self" has formed which is then disrupted. The stressor that adolescent participants were responding to may not have been the disruption to self-concept, but the disruption to a perceived life trajectory such as gaining education qualifications and transition to work. Such a disruption could have ongoing impacts on identity into adulthood, reflecting similar concerns examined in a cohort of young adults with cancer (Grinyer, 2007).

Clinical implications

The findings have clinical implications for individuals, families and schools. There are also wider implications based on the need to address the limited understanding and misrepresentations of TS at a societal level. There is strong evidence to support the use of behavioural therapies in treating individuals with TS (for example, Wile & Pringsheim, 2013). One such approach, habit reversal treatment, involves increasing awareness of the premonitory urge and learning to perform a competing action which is incompatible with performing the tic (Azrin & Nunn, 1973). The evidence in this review suggests it could also be beneficial to integrate elements of other therapeutic approaches that can help address the psychological impact of stigma and an acceptance of the condition in relation to self-identity.

Acceptance and commitment therapy (ACT: Hayes et al., 1999) could help individuals to accept changes that TS brings and to enable identity continuity through the identification of underlying values (Chan, 2013). ACT uses techniques such as diffusion and mindfulness to help individuals detach from difficult thoughts and develop flexible ways of coping (Hayes & Storsahl, 2011). Although there is little evidence for the efficacy of ACT in TS populations,

there is evidence that ACT can be effective in neurological conditions such as Parkinson's disease where interventions are associated with reduced distress and increased psychological flexibility (Hill et al., 2017). Any individual therapeutic protocols for use with people with TS would need to consider the commonly co-occurring neurodevelopmental issues, such as difficulties with attention, in order to help manage the cognitive demands required when engaging with such an intervention.

Compassion-focused therapy (CFT) could be another complementary approach to the use of behavioural therapies, especially given its biopsychosocial foundations (Gilbert, 2009). CFT encourages the development of a compassionate mind which can be applied to the self and other people to reduce distress (Gilbert, 2014). The approach can be particularly effective in addressing feelings of shame and guilt by fostering an acceptance of difficult emotional states (Leaviss & Uttley, 2015). This could be particularly beneficial to parents of children with TS. Although limited evidence exists on the use of CFT with this population, the approach has been found to reduce anxiety and depression in the mothers of children with cerebral palsy (Khoshvaght et al., 2021).

However, it is important to consider the wider societal context when aiming to improve coping with a stigmatised condition. A systematic review of interventions for reducing health related stigma found interventions should target multiple levels; interpersonal, organisation, community and government levels (Heijnders & Van Der Meij, 2006). Improving awareness of TS at a society level must be conducted with sensitivity and accuracy, as there is a risk that increased media depiction of an inaccurate TS portrayal can strengthen misperceptions and stigmatising beliefs (Cox et al., 2022). The trivialisation of TS by others and the role of humour in perpetuating TS stigma suggests a new understanding of TS should be developed (Malli &

Forrester-Jones, 2019). One potential approach is through photovoice, an ethnographic and anthropologic way of encouraging activism within stigmatised groups by using photography (Catalani & Minkler, 2010). Through publication of images alongside personally meaningful narratives, this intervention has been found to reduce internalised stigma in addition to challenging stereotypes held by non-stigmatised viewers (Russinova et al., 2014). A new understanding of TS could become more like that of autistic spectrum condition, which is seen as a mix of neurodevelopmental factors combined with social expectations (Bervoet et al., 2023). Thinking of TS as a Tourettic experience could broaden general public understanding of the condition into one that is more than just tics (Bervoet et al., 2023).

Limitations and future research

Although the meta-ethnographic approach allowed for the participants' experiences within the studies to be preserved (Britten et al., 2002), the large number of studies included meant it was difficult to capture all the nuance within the themes. For instance, there may have been subtle differences in experience among the different ages included. This is particularly important given the impact that age could have on development of identity (Erikson, 1968). Parent voices were heard, but there was no inclusion of siblings or partners. Future work could focus on wider family experiences, including the partners of adults with TS. The studies were weighted towards the child population, with fewer studies looking at the experience of adults. Although the effects of stigma have been found to be similar across the lifespan (Smith et al., 2015), there could be differences in childhood experience, for example, parenting styles, that have consequences for adult experience. Further exploration of adult coping would be an important avenue for research. This could lead to improved psychoeducation for adults, for example, with information on managing tics at work and education for employers.

The findings were also biased towards the experiences of people from English speaking, Western countries. Cultural factors may also have an impact on the presentation of TS symptoms and in the distress caused by the symptoms (Robertson et al., 2009). Given that cultural factors have an influence on the nature of stigma (Abdullah & Brown, 2011), it would be interesting to understand nuances that might arise by looking at coping with the stigma of TS across different cultures. The findings were also limited in that the studies included did not consider demographic factors of participants, such as ethnicity and socioeconomic status. These factors give rise to social identities in addition to a diagnosis of TS. The multiple identities arising from social constructs interact to influence access to social capital, and therefore, to coping resources (Institute of Medicine, 2011). Individuals with multiple devalued social identities experience greater oppression (Institute of Medicine, 2011). For example, individuals with HIV are subject to increased stigma if they also belong to minoritized ethnic, gender and sexuality groups (Earnshaw et al., 2015; Rao et al., 2008). Further research on TS taking an intersectionality approach (for example, see Cole, 2009) would be useful.

The variability of the papers included, as evaluated by the CASP tool (2018), may have impacted on the findings. Few papers acknowledged the researchers' potential influence on their data which may have impacted the validity of the data. Future research in this area could benefit from more explicit discussion of researcher reflexivity to enhance credibility of the findings (Yardley, 2000). However, meta-synthesis methods can guard against this limitation by comparing data from through multiple data sources and independent reviewers (Walsh & Downe, 2005).

The papers included were not aiming to address coping with TS directly, rather this was inferred from first hand experiences of living with the condition. This may be why there was no mention of medication in any of the studies. It would therefore be useful to explore the concept of coping more directly with this population. The findings from this research would be a helpful starting point to ensure that future research, whether quantitative or qualitative, when looking at coping includes the psychosocial as well as the biological nature of coping. Another limitation is that the papers included in this review do not determine the nuance of how co-occurring conditions impact on TS. The samples in the studies were not differentiated along these lines. Given that the quantitative research suggests co-occurring conditions such as ADHD and OCD can cause as much or even more distress than the tics themselves (Bernard et al., 2009), it would be useful to understand this from a qualitative perspective.

Conclusion

This review has identified new findings that contribute to the existing literature exploring how individuals and parents cope with TS. Findings indicate that coping involves the need to integrate TS with identity, to exert control over tics and to challenge the misrepresentations of TS in wider society. A supportive environment provided by parents and friends enables individuals to feel proud that they can control their tics, and this allows for the positive integration of TS into identity. Implications for clinical treatment and wider social recommendations have been discussed, including the enhancement of education interventions. Greater understanding of how individuals and those around them cope with the often painful and exhausting condition that is TS will help to maximise wellbeing.

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Figure 1 – A: PRISMA flow diagram

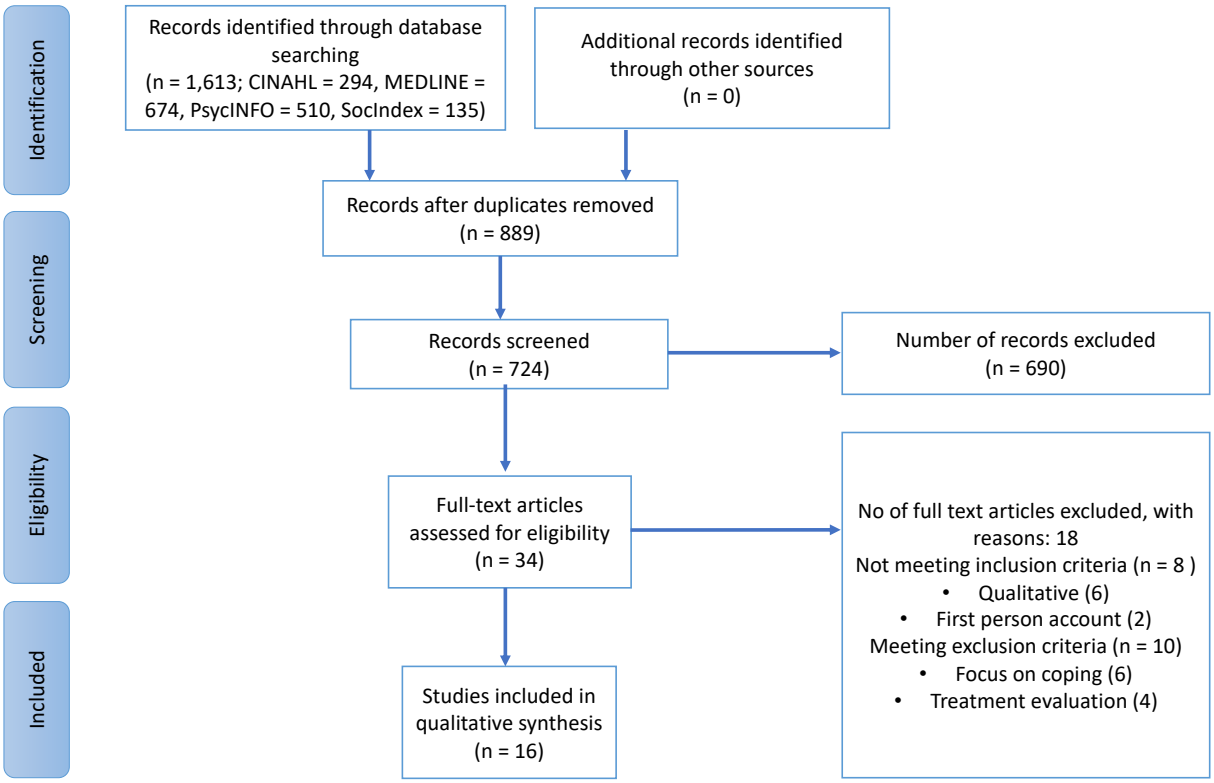

Figure 1 – B: Conceptualisation of how themes fit together

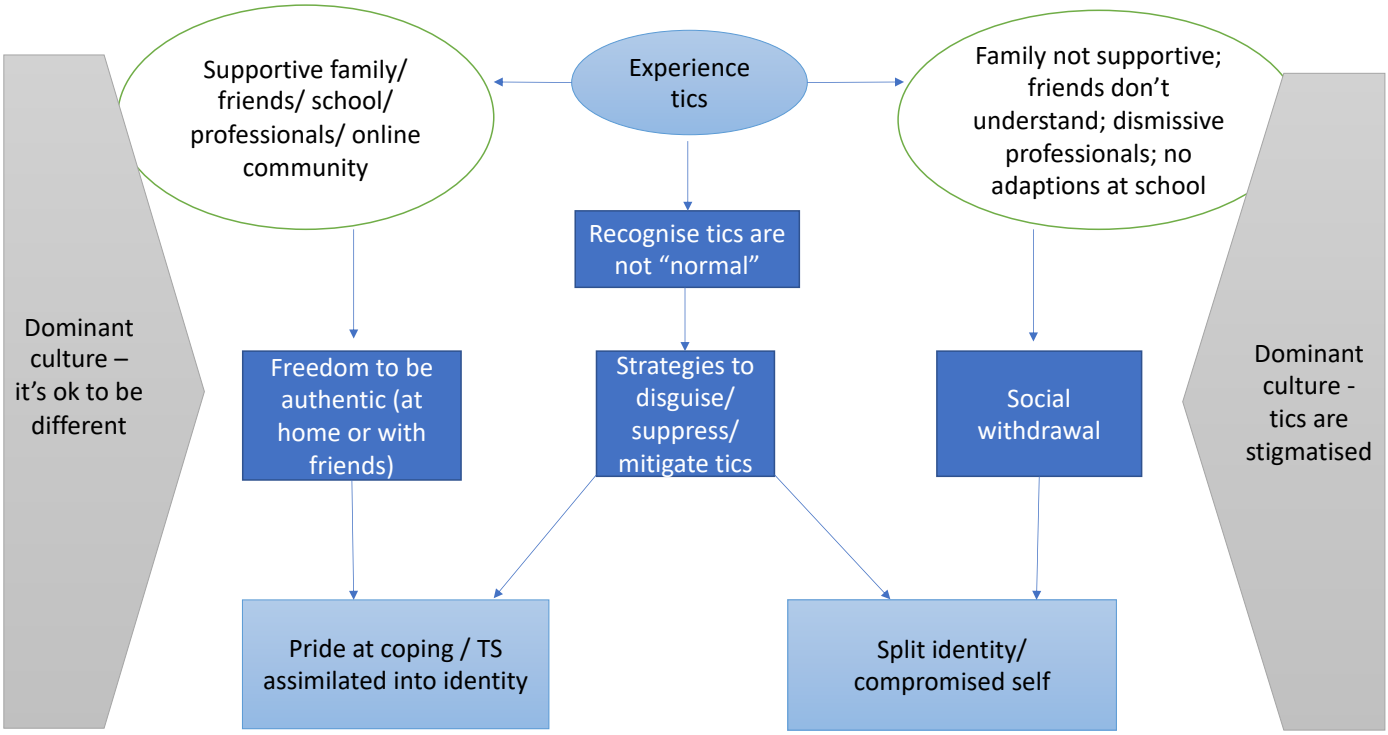

Table 1 - A: Search strategy and database search terms

CINAHL [294 hits]	
Focus 1 **Tourette's syndrome**	**Focus 2** **Coping**
MH "Tourette Syndrome" MH "tourette" Free text **"***Tourette**** OR *tics* OR *"tic disorder""*	MH "Coping+" MH "coping" MH "Family Coping" MH "Social Adjustment" MH "Adaptation, Psychological+" Free text "*Coping* OR *adjustment* OR *wellbeing* OR *experience* OR *"living with""*
MEDLINE [674 hits]	
Focus 1 **Tourette's syndrome**	**Focus 2** **Coping**
MH "Tourette Syndrome" Free text **"***Tourette**** OR *tics* OR *"tic disorder""*	MH "Adaptation, Psychological+" MH "coping" MH "Social Support" MH "Emotional Regulation" MH "Cognitive Reserve" Free text "*Coping* OR *adjustment* OR *wellbeing* OR *experience* OR *"living with""*
PsychInfo [510 hits]	
Focus 1 **Tourette's syndrome**	**Focus 2** **Coping**

DE "Tourette Syndrome" DE "Tic Disorders" DE "Tics" Free text **"***Tourette** OR *tics* OR *"tic disorder""*	DE "Coping" DE "Adjustment" DE "Codependency" DE "Coping Style" DE "Adaptability (Personality)" DE "Emotional Adjustment" DE "Emotional Control" DE "Identity Crisis" DE "Emotional Control" DE "Anger Control" DE "Emotional Exhaustion" DE "Emotional Processing" DE "Sense of Coherence" DE "Stress and Coping Measures" DE "Coping Behavi*" DE "Coping Style" DE "Hopelessness" DE "Internal External Locus of Control" DE "Health Locus of Control" DE "Interpersonal Control" DE "Abuse of Power" DE "Oppression" DE "Self-Control" DE "Delay Discounting" DE "Delay of Gratification"

	DE "Self-Determination" DE "Helplessness" DE "Learned Helplessness" DE "Self-Confidence" DE "Self-Stigma" DE "Psychological Capital" DE "Resilience (Psychological)" DE "Self- Efficacy" Free text **"***Coping* OR *adjustment* OR *wellbeing* OR *experience* OR *"living with""*
SocIndex [135 hits]	
Focus 1 **Tourette's syndrome**	**Focus 2** **Coping**
Free text **"***Tourette** OR *tics* OR *"tic disorder""*	SU "ADAPTABILITY (Personality)" SU "PSYCHOLOGICAL well-being" SU "SOCIAL adjustment" Free text **"***Coping* OR *adjustment* OR *wellbeing* OR *experience* OR *"living with""*

Table 1 – B: Characteristics of the studies included for synthesis

Author(s) and title	Country	Research question/aim(s)	Sample	Methodology	Findings related to coping
Buckser (2006) The empty gesture: Tourette syndrome and the semantic dimension of illness	USA	To explore challenges in the classification of illness and TS	Adults: 'About two dozen' (no further information) and participant observations at TS support groups and camps	Interview: Ethnography	• Control and uncontrol – helping to make TS understandable to others • The physical and the mental – certain emotional states can help tics recede

Buckser (2008) Before your very eyes: illness, agency, and the management of Tourette Syndrome	USA	To explore effects of disease on social experience and effects of culture on disease	16 adults: 11 men, 5 women, age 21 –62	Interview: Ethnography	• Displacement – creating time and space to tic • Misattribution - putting tics in other categories of behaviour • Contextualization - putting tics into a discourse
Congdon (2014) What's wrong with Me?: An Autoethnographic Investigation of the Co-Cultural Communicative Practices of Living with Tourette Syndrome during Adolescence	UK	To explore living with TS as it relates to education	1 adult with TS (the researcher)	Autoethnographic methodology	• Non-assertive assimilation: negotiating with relationships of authority • Aggressive assimilation: negotiating relationships with peers • Non-assertive separation: the convergence of negotiating relationships of authority and with peers

Cutler et al. (2009) The quality of life of young people with Tourette syndrome	UK	To explore the effect of TS on young people's quality of life	11 young people, 8 male, 3 female, age 8–17	Mixed methods: Focus groups and thematic analysis	• Needing to control tics • Integrating TS into self-identity
Edwards et al. (2017) A Qualitative Exploration of the Experiences of Children and Adolescents with Tourette Syndrome	Canada	To explore the experiences of youth with TS	13 young people, 6 – 17 years old	Semi-structured interviews and two questionnaires. Thematic analysis.	• Coping with TS – control symptoms including ignoring, suppressing, or disguising tics.
Lee et al. (2016)	Taiwan	To explore self-experience in	12 adolescents	Phenomenological	• The secular 'me' from transmigration • Peer recognition • Opportunity for self-identity

'Living with tics': self-experience of adolescents with Tourette syndrome during peer interaction		the context of peer interactions			• Adjustment to symptom-related situations • Endeavouring to maintain the image of normalcy
Lee et al. (2018) Social adjustment experiences of adolescents with Tourette syndrome	Taiwan	To explore experience of social adjustment of adolescents	16 adolescents	Descriptive phenomenological approach	• Compromising oneself to integrate into society • Conflict between autonomy and authority • Helping factors in developing self-identity • Two-faced – comparing to people who are worse off • The power of accepting that TS is a part of you
Lludlow et al. (2018)	UK	To explore parents' experiences, the	15 parents	Thematic analysis	• Coping with children's challenging behaviours

A qualitative exploration of the daily experiences and challenges faced by parents and caregivers of children with Tourette's syndrome.		challenges they face and the support mechanisms they have found to be most helpful			• Addressing misconceptions and lack of understanding of professionals and the lay public • Addressing negative experiences of their children's education • Support and services for families with TS
Malli et al. (2019) "Tourette's Is a Lonely Place": an Interpretative Phenomenological Analysis of the Personal Experience and Identity of Adults with Tourette's Syndrome	UK	To evaluate the social and personal cost of living with TS during adulthood. To explore self-identity threat,	16 adults	Interpretative phenomenological analysis	• Living with Tourette's Syndrome • Supportive Relationships • Involuntary Isolation • Social Identity Threat and Social Withdraw • Lack of Resources for Adults with Tourette's Syndrome – wanting to belong to a TS community group

		social withdraw, and self-stigma			
Malli & Forrester-Jones (2022) Stigma and Adults with Tourette's Syndrome: "Never Laugh at Other People's Disabilities, Unless they have Tourette's—Because How Can You Not?"	UK	To explore experiences of stigma	20 adults, 14 males, 6 females	Mixed methods: Thematic analysis	• Stigma management – acting as a TS educator to others and adapting to the world around them
O'Connor et al. (2009)	Canada	To explore representations of physical,	3 adults: female – age 26, males – age 29, 55	Interview: Phenomenological method	• Maintaining vigilance and prudence around others

'I'm cured but..': Perceptions of illness following treatment		psychological/ psychophysiological illness Explores three types of health condition: one physical (heart transplant), one psychological (panic disorder) and the third psychophysiological (TS)			• Hope that others would see them for who they are • Develop strategies to foresee the reactions of others • Developing ambivalence – take it or leave it approach

O'Hare et al. (2017) Youth with Tourette syndrome: Parental perceptions and experiences in the Australian context	Australia	To enhance understandings of the impact of TS on the parents of diagnosed young people	22 mothers of young people with TS	Semi-structured interviews conducted ss part of a larger qualitative and quantitative community-based study. Inductive content analysis	• Mother forced to take charge, advocate, become the expert • 'Living worried' and 'Staying in the struggle • 'Tangled' – trying to disentangle TS and associated neurodevelopmental conditions • Critical times – preparing for difficult transitions and situations • The diagnostic experience – educating healthcare professionals • Grief and loss of the 'ideal child' • Family first—home as sanctuary • Bridge to the outside world – using support groups • School as a key player

Pine et al. (2022) Perceptions of Parents and Caregivers in New Zealand: Educational Experiences of their Children with Tourette Syndrome	New Zealand	To explore parents' and caregivers' attitudes and experiences of education for their child diagnosed with TS	75 parents and caregivers of young people aged 6 to 18	Thematic analysis	• Accommodations made by school to support children with TS • Separate space for children to tic away from their peers • Calls for increased education about TS for teachers and peers
Smith et al. (2016) Investigating young people's experiences of successful or helpful psychological interventions for tic	UK	To explore experiences of having received a helpful psychological intervention in	7 young people aged 10–17 years, 2 female, 5 males	Interpretative phenomenological analysis	• The challenging battle with TS and the sense of self • Making sense of the lived experience • Useful strategies to manage experience • The spectrum of positive change

disorders: An Interpretative Phenomenological Analysis study		relation to their tics			
Travis & Juarez-Paz (2020) Experiences of Tourette Syndrome Caregivers With Supportive Communication	USA	To understand TS caregivers' experience with supportive communication	11 carers	Semi-structured interviews; Grounded theory	• Struggling Is the New Normal (learning to accept difficulties) • The Validated Caregiver – receiving encouragement from other parents • The Isolated Caregiver - supportive communication that questions the participants' experiences is isolating
Wadman, et al. (2013) 'Everybody just thinks I'm weird': A qualitative exploration of the	UK	To explore the psychosocial impact of TS	6 young people: 4 male, 2 female age 14 –16	Semi-structured interview: Interpretative	• Learning to cope well with TS • Developing supportive friendships • Talking to peers about TS

psychosocial experiences of adolescents with Tourette syndrome				phenomenological analysis	

Table 1 - C: Results of critical appraisal using CASP Qualitative Checklist

Paper	Rank	Clear statement of aims	Qualitative	Research design	Recruitment strategy	Data collection	Role of researcher	Consideration of ethical	Sufficiently rigorous	Clear findings	How valuable is research	Total score
Buckser (2007)	14th	Y	Y	2	1	1	1	2	2	3	3	15
Buckser (2008)	11th	Y	Y	3	1	2	1	2	2	3	3	17
Congdon (2014)	12th	Y	Y	2	3	1	1	2	1	3	3	16
Cutler et al (2009)	14th	Y	D	3	3	3	1	2	1	1	1	15
Edwards et al (2017)	3rd	Y	Y	3	2	3	1	3	3	3	3	21
Lee et al (2016)	9th	Y	Y	2	3	3	3	3	1	3	1	19
Lee et al (2018)	3rd	Y	Y	2	3	3	3	3	2	2	3	21

Ludlow (2018)	6th	Y	Y	3	3	3	1	3	3	3	1	20
O'Connor (2009)	14th	Y	Y	2	2	3	1	3	1	3	1	15
O'Hare (2006)	3rd	Y	D	2	3	3	1	3	3	3	3	21
Malli et al (2019)	1st	Y	Y	1	3	3	3	3	3	3	3	22
Malli & Forrester-Jones (2022)	10th	Y	D	2	3	2	1	3	1	3	3	18
Pine et al (2022)	12th	y	y	3	1	2	1	3	2	3	3	16
Smith et al (2016)	6th	Y	Y	2	3	3	1	3	2	3	3	20
Travis & Juarez-Paz (2006)	6th	Y	Y	2	3	3	1	2	3	3	3	20

Wadman et al. (2013)	1st	Y	Y	3	1	3	3	3	3	3	3	22

Table 1 – D: Table showing how themes emerged from concepts

Initial concept identified	**Themes emerging from concepts**	**Refining of themes**	**Final theme**	**Relevant studies**
TS clearly visible, ever present. Part of self or external to self. Living with vs living through, that is, those that accept it as part of identity and those that fight against it. Because of the reaction of others, they are aware of their tics. Social perceptions are internalised. Beyond stopping tics. Acceptance that tics will always be there. Can grow in strength if you see TS as a friend and learn from it. Hope that others would see them for who they are not the tics. Having to negotiate new identity with TS. Integrate TS into their sense of selves – process helped by diagnosis and friends. Wanting others to understand the impact of tics. Wanted to know they were not the only ones. Feeling welcomed strengthens self-confidence and self-identity.	How TS relates to an individual and family member's personal and social identity Supportive friendships can help integrate TS into identity. Accepting the person means accepting their tics. Validation from others about how difficult it is living with TS – a daily	Becoming comfortable with TS as either part of self or separate to self can reduce the negative emotional impact of TS. For individuals being themselves accepted by supportive friends and family aides this process. For family members, this process	Redefining the self and social identity	Wadman et al (2013), Malli et al (2019), Lee et al (2018), Smith et al (2016), Lee et al (2016) O'Connor et al (2009), Congdon (2014), Malli & Forrester-

Therapy groups were normalising. Tolerance and respect of parents helps to free them from TS. Inspired by others who have TS. If peers interact normally TS is not a barrier – may even forget it exists. Struggling is the new normal for parents. Need to grieve for lost child and come to terms with it. Struggle leads to shift in expectations. Tangled – where is the line between TS and normal adolescent behaviour? Some parents wanted support groups, some not. Parents who concentrated on their child's achievements and distilling confidence in their child, also reported having an easier time. Validation important because of feelings of parent guilt.	struggle – helps individuals and family members to feel supported.	of acceptance is aided by feeling validated by others as to how difficult it is to live with TS.		Jones (2022), Cutler et al (2009), Ludlow et al (2018), Travis & Juarez-Paz (2006), O'Hare et al (2006), Pine et al (2022)
Responsibility to hide tics. Pride when controlling tics. Ignoring, supressing or disguising tics. Choose between trying to conceal tics or exposing their true self. Learning to avoid mood and physical states that trigger tics. Control	Measures are taken to control the tics, both physically and mentally.	Concealment of tics is a mechanism commonly utilised by individuals to manage	Controlling the visible presentation of TS	Wadman et al (2013), Malli et al (2019), Edwards et al

over tics has important meaning. Endeavor to appear 'normal' means feeling pain to suppress tics. Confidence to tic in public grows with age. Tics a matter of self-presentation - displacement, misattribution, and contextualization. Sometimes need isolation to get tics out of system. Ambiguity of whether individuals can control their tics or not. Supressing tics, hoping to assimilate into the norm. Controlling tics is physically and emotionally exhausting. Need to supress tics is all consuming. Practicalities of coping – eating meals earlier in the day to avoid when tics get worse in evening. Going on holiday to quiet places. Family first – respond to social isolation by creating sanctuary at home for child to let it all hang out. Home as a safe space.	Controlling tics can mean physically removing themselves or their children with TS from social spaces leading to social withdrawal Freedom to tic at home means home becomes a safe haven Parents are also involved in the control of their children's tics – for presentation and behavioural reasons	their or their family's "spoiled identity".		(2016), Lee et al (2018), Smith et al (2016), Lee et al (2016), O'Connor et al (2009), Buckser (2008), Buckser (2006), Malli & Forrester-Jones (2022), Cutler et al (2009)

	Individuals can learn to coexist with TS, removing the need to control tics, by learning to cope with other people's judgement			
Lack of self-advocacy groups in relation to TS. Protest against discrimination is still individual and disorganised. Importance of non-judgemental friends and partners. importance of explaining the involuntary nature of TS. Friends can be more sympathetic if they think it's medical. Are tics medical or mental? More power in the medical	Educating others about TS is a way of coping with feeling dismissed and misunderstood Feeling understood reduces the shame	The worst thing is misunderstanding. Therefore, coping requires education of those around the person with TS.	Challenging the narrative	Wadman et al (2013), Malli et al (2019), Edwards et al (2016), Lee et al (2018),

model. Semantic difficulties reflect cultural views of body and illness. Educating others about medical causes of TS. Making efforts to understand and educate selves about TS. Understanding makes lives easier. Having to educate other people. Feeling isolated by people not accepting and misunderstanding. Disbelief and dismissal. Battling ignorance, fighting for understanding and acceptance. Mother forced to take charge, become an advocate and expert. Support groups act as a bridge to the outside world. People who get it. Counter to distress from isolation and misunderstanding. Parents want more education so people know tics aren't just swearing. Strategies of displacement, misattribution, and contextualization strongly affect both the symptoms themselves and the subjective experience of the illness. They also affect the perception of TS in the	associated with a stigmatised condition Given cultural dominance of medical model, it is more acceptable to explain TS as a medical issue. Ability to control tics means TS doesn't sit entirely in the medical model More organised advocacy could help at a population level	Adaptions at school need to be made therefore coping requires open communication and understanding between family and school. It is important that people feel they have a sense of agency and control in their environment.		Smith et al (2016), Lee et al (2016), O'Connor et al (2009), Congdon (2014), Buckser (2008), Buckser (2006), Malli & Forrester-Jones (2022), Cutler et al (2009),

larger culture. Mid adolescents can cope with TS if they have ways of explaining their tics to peers. The findings show the impact of local culture on the social adjustment processes for adolescents with TS. No longer willing to comply with parents and experts who don't have TS. Autonomy leads to better coping strategies. Education of teachers and support staff. School were able to balance providing extra provision without making child look different. Having a card to hold up if they needed to leave the classroom. Having to fight to get support from professionals. Dealing with misconceptions and feeling dismissed by teachers. Validation from professionals. School is critical. Diagnosis as critical time. Having to educate professionals. But diagnosis can give relief. Accommodations made by school eg sensory toys, extra time, taking exams in private room, supported by reader,	Conflict between authority and autonomy Importance of school Getting the right support from professionals			Ludlow et al (2018), Travis & Juarez-Paz (2006), O'Hare et al (2006), Pine et al (2022)

supportive teacher or peer. Passes and cards. Strategies to manage disruptive tics. Working collaboratively between teacher and schools. Especially role of private space to tic – walk around, be on own and reduce anxiety will reduce tics.				

Appendix 1 – A: Synthesis of concepts to develop the final themes.

Study	Theme 1: Redefining the self and social identity	Theme 2: Controlling the body	Theme 3: Challenging the narrative
Buckser (2007)			• Ambiguity of whether people with TS can control their tics or not. This leads to misattribution errors and for people being held responsible for tics • Are tics medical or mental? More power in the medical model • Semantic difficulties reflect cultural views of body and illness
Buckser (2008)		• Tics a matter of self-presentation. displacement, misattribution, and contextualization	• Can portray tics as medical condition or disability. Agency shaped by dominant illness perception in culture

		• Sometimes need isolation to get tics out of system	• Friends can be more sympathetic if they think TS is a medical, not a mental health, condition
Congdon (2014)	• Having to negotiate new identity. Author had not been born into a marginalised group, so had to learn new strategies to cope	• Mirroring, dissociating, strategic distancing when communicating with peers	
Cutler et al (2009)	• Some managed to integrate TS into their sense of selves. Process over time that was helped by diagnosis and family and friends' acceptance	• Need to supress tics to fit in and avoid attention which was perceived as negative. All consuming	
Edwards et al (2017)	• Desire to belong to a TS group as they wanted to know they were not the only ones	• Ignoring, supressing or disguising tics. This varies in how helpful it is • Home as safe place. Don't need to manage tics. Holding tics in in public vs letting everything out at home	• Importance of explaining the involuntary nature of TS symptoms to peers.

Lee et al (2016)	• Can grow in strength if you see TS as a friend and learn from it. • Important to have developed sense of who you are first before you can win affection from peers • Tolerance and respect of parents helps to free them from TS • Supportive friends, parents and teachers inspire the courage of self-acceptance	• Learning to avoid mood and physical states that trigger tics • Endeavor to appear 'normal' means feeling pain to suppress tics • As participants with TS grew older, they developed confidence to be with their tics in public, and to stop caring what others think • No need to control tics – can even forget that they are there is peers interact normally	• Inspired by others who have TS – importance of wider narrative of TS for adolescents with TS
Lee et al (2018)	• Because of the reaction of others, they are aware of their tics • Power of accepting tics are part of who you are	• Must choose between trying to conceal tics or exposing their true self • Many choose to hide their tics	

	• TS gives strength • TS is a stepping-stone to being courageous and mature • TS has led to more empathy for others • No longer willing to comply with parents and experts who don't have TS. Autonomy leads to better coping strategies • Feeling welcomed by friends - despite TS - strengthens self-confidence and self-identity		
Ludlow et al. (2018)	• Some wanted support groups, some not • Parents who concentrated on their child's achievements and distilling	• Practicalities of coping – eating meals earlier in the day to avoid when tics get worse in evening. For example, going on holiday to quiet places	• Having to fight to get support from professionals • Fighting misconceptions

	confidence in their child reported having an easier time	• School were able to balance providing extra provision without making child look different. • Having a card to hold up if they needed to leave the classroom to tic in private	
O'Connor et al.(2009)	• Hope that others would see them for who they are not the tics	• Interventions to focus on control through relaxation instead of control of other's perception of self	
O'Hare et al. (2006)	• Requires self-sacrifice and determination • Tangled – where is the line between TS and normal adolescent behaviour? • Grief and loss of the ideal child. At the same time guilt because TS is not life threatening.	• Mother takes on day to day care. Staying with the struggle • Family first – respond to social isolation by creating sanctuary at home for child to let it all hang out	• Battling ignorance, fighting for understanding and acceptance. Mother forced to take charge, become an advocate and expert • Support groups act as a bridge to the outside world. People who get it.

			Counter to distress from isolation and misunderstanding • Having to educate professionals. But diagnosis can give relief
Malli et al. (2019)	• Two distinct and divergent groups. Those that accepted their condition as part of their own identity and life and were willing to work with it as opposed to against it; and those that felt inferior due to TS and were grieving the loss of normalcy. • Living with vs living through. • In both groups, the participants did not view TS as a dominant identity characteristic.	• 'Living through TS' group have the attitude that they don't want TS so have to control it. • Avoidance of social interactions rather than tackle anxiety. TS as a "lonely place".	• There is a lack of self-advocacy groups in relation to TS, and protest against discrimination is still individual and disorganised.

	• None of the participants in the group tried to counteract the discrimination of TS by increasing identification with the TS group. But could have benefited from adult support group. • Supportive relationships. Non-judgemental friends. Support of partner.		
Malli & Forrester-Jones (2022)		• Supressing tics, hoping to assimilate into the norm • Supressing tics is important despite being physically and emotionally exhausting	• The role of Tourette's educator became all consuming – so they could not be themselves in public • Educating others about medical causes of TS
Pine et al. (2022)		• Home as a safe space • Accommodations made by school eg sensory toys, extra time, taking	• Parents want more education so people know tics aren't just swearing

		exams in private room, supported by reader, supportive teacher or peer. Passes and cards. • Strategies to manage disruptive tics. Working collaboratively between teacher and schools. Especially role of private space to tic – walk around, be on own and reduce anxiety will reduce tics	• Education of teachers and support staff.
Smith et al (2016)	• Battle between TS and self • TS as external to self – seen as an irritating, constant presence • Social perceptions of TS internalised to develop self-identity • Therapy helped synthesise identity with TS. Some integrated TS into	• Strategies to control tics were highly valued • Internal techniques to manage tics – for example, managing mood helps manage the tics • Control over tics has important meaning	

	identity, whereas some talked about regaining control and self-esteem without the need to integrate TS to self-identity • Therapy groups were normalising • Beyond stopping tics. Need to develop acceptance that tics will always be there		
Travis & Juarez-Paz (2006)	• Struggling is the new normal. • Need to grieve and come to terms with having a child with TS • Struggle leads to shift in expectations • Acknowledgement, connection, answers by other parents, professionals and children		• Making efforts to understand and educate selves about TS. Understanding makes lives easier • For others to understand the exhaustion and attribution to neurological condition

	• Validation important because of feelings of guilt		• Having to educate other people otherwise feel isolated by people not accepting and misunderstanding
Wadman et al. (2013)	• TS clearly visible, ever present • TS is part of self • Or TS is external to self • Wanting others to understand the impact of tics • Friends who are accepting	• Feeling a responsibility to hide tics • Feeling pride when learnt to control tics in public	

Appendix 1 – B: Journal of Health Psychology: Instructions for authors

Manuscript Submission Guidelines: Journal of Health Psychology

This Journal is a member of the Committee on Publication Ethics

Please read the guidelines below then visit the Journal's submission site http://mc.manuscriptcentral.com/jhealthpsychology to upload your manuscript.

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1. What do we publish?
   1.1 Aims & Scope
   1.2 Article types
   1.3 Writing your paper
2. Editorial policies
   2.1 Peer review policy

1. What do we publish?

1.1 Aims & Scope

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The Editorial Board of the Journal of Health Psychology considers for publication:

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(d) Open peer commentaries on recent articles in this journal or topical issues (up to 2,000 words counting 500 words per table and figure).

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2. Editorial policies

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Section Two: Research Paper

Navigating the social world with neck dystonia – An interpretative phenomenological analysis

Word Count – 7,693
(excluding title page, references, figures, tables and appendix)

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Prepared for: Journal of Health Psychology

Abstract

This research aimed to provide insight into how people with neck dystonia navigate the social world. Neck dystonia is a neurological condition, characterised by involuntary movements of the neck muscles, causing twisted head positions and often pain and head tremor. Ten participants with neck dystonia were interviewed and the data was analysed using an interpretative phenomenological analysis approach. Three themes were constructed: (1) dismissed by others for having an unfamiliar condition; (2) negotiating a new social identity; and (3) managing the stigma of a visible condition. It is proposed that psychological support could be beneficial to people with neck dystonia who experience difficulties arising from identity management and stigma. Systemic interventions, such as education campaigns, are also proposed to help address stigmatising attitudes. Future research is suggested that would contribute to a deeper understanding of distress in this population.

Keywords: adults, neck dystonia, cervical dystonia, spasmodic torticollis, social world, social support, stigma, interpretative phenomenological analysis, qualitative

Introduction

Dystonia refers to a group of neurological conditions characterised by sustained or intermittent muscle contractions and spasms (Albanese et al., 2013). These uncontrolled movements can be repetitive and can cause twisting, often painful postures (Werle et al., 2014). Dystonia is the third most common movement disorder, after idiopathic Parkinson's disease and essential tremor (Defazio, 2010). Prevalence is estimated at 16.43 per 100,000 of the

population (Steeves et al., 2012). However, this could be an underestimate as the condition can be difficult to diagnose (Albanese et al., 2019). The classification of dystonia involves two major axes (Albanese et al., 2019). The first axis includes age of onset and body distribution. The second axis involves aetiological factors such as whether dystonia is inherited, with a known genetic link, acquired through, for example, infection or toxicity, or idiopathic, with no known cause. Dystonia can be isolated or combined with other conditions such as Parkinson's disease and cerebral palsy (Albanese et al., 2019).

Adult-onset, idiopathic, focal dystonia (affecting specific body parts) is the most prevalent form of dystonia (Defazio et al., 2013). Neck dystonia, also known as cervical dystonia or spasmodic torticollis, is the most common form of focal dystonia with an estimated prevalence of 1.07 per 100,000 (Steeves et al., 2012). It is characterised by involuntary movement of the neck muscles, leading to twisted head postures, and can be accompanied with pain and tremor (Defazio et al., 2013; Kuyper et al., 2011). Treatment for neck dystonia is focused on reducing motor symptoms and often involves regular injections of botulinum toxin into the affected muscles (Albanese et al., 2019). Onset of neck dystonia is usually between the ages of 30 and 50 years old (Carvalho Aguiar & Ozelius, 2002) meaning many people experience diagnosis when they are working and/or caring for family members (Skogseid et al., 2005). Women are affected 1.5 to 1.9 times more often than men (Warner, 1999). Neck dystonia is a chronic condition, although 10 to 20 per cent of people may experience a temporary remission (Dauer et al., 1998).

In addition to motor symptoms, people with neck dystonia also commonly experience a range of psychological difficulties including poor sleep, sensory issues, anxiety and depression (Smit et al., 2017; Stamelou et al., 2012; Kuyper et al., 2011). These difficulties

have been shown to have a greater impact on health-related quality of life than motor symptoms (Ndukwe et al., 2020; Klingelhoefer et al., 2021). This has led to debates as to whether psychological distress is a primary feature of dystonia (Ndukwe et al., 2020), or a consequence of living with this chronic condition (Comella & Bhatia, 2015; Lewis et al., 2008)

Given the complexity and interrelatedness of these variables, a biopsychosocial framework (Engel, 1977; Wade & Halligan, 2017) would be an appropriate way to understand the condition and the impact on individuals. This framework regards health conditions as a complex interaction of biological, psychological, and social factors (Engel, 1977). Social factors include interpersonal relationships, family environment, social support and isolation, social expectations, cultural factors, and work history (Gatchel et al. 2020). The biological component refers to the physical aspects of ill-health, such as disease, genetics, and the impact of medication, whereas the psychological component refers to emotional, cognitive and behavioural aspects of ill-health, such as personality traits and coping strategies (Vögele, 2015). The social world is particularly important when considering chronic health conditions which do not have an identified cure (Bury, 1991), as is the case for dystonia. In the social model of disability, the social world and specific chronic illnesses interact to create an illness experience (Oliver, 1996). This experience can be more complex than the pain and fatigue of physical symptoms (Conrad, 1990). For instance, when one family member is ill the whole family can be disrupted, and this disruption can be the most meaningful element of illness experience (Leahey & Wright, 1987).

Recognising this social component has led to important developments in understanding the disadvantages that people face when living with chronic ill health. These disadvantages have been referred to as disablism and this can be experienced as two separate forms of

oppression (Thomas, 2007). Structural oppression involves barriers outside the individual, for example, physical and social exclusion, that result from an individual's impairment. Psycho-emotional disablism refers to oppression relating to an individual's emotional wellbeing, impacting self-esteem and self-confidence (Thomas, 2007; Reeve, 2006). Other people can cause this oppression through invalidating actions, for example, staring at people with visible differences, telling jokes and making thoughtless comments (Reeve, 2006). However, oppression is not an inevitable consequence and can depend on other elements of identity, for example, age, ethnicity, gender and social class (Liasidou, 2013). People with chronic health conditions may need to overcome structural and psycho-emotional oppression in addition to easing the physical symptoms (Shakespeare, 2013). This could be the case for people with neck dystonia, whose quality of life has been found to be reduced by pain and disability, but also social and emotional factors (Tomic et al., 2016), including factors which relate to disablism such as self-esteem and social participation (Ben-Shlomo et al., 2002).

Other people also have a role in defining what is 'normal' and who belongs to which social groups. Goffman (1963) described a stigmatised individual or group as possessing features which differ from a social norm, either due to physical differences, 'character flaws' or identification with a particular group on the grounds of, for example, race or religion. One study found 51% of participants with neck dystonia reported the feeling of stigmatisation and this was particularly prevalent for those under 60 years (Klingelhoefer et al., 2020). This supports the findings of an earlier study which found people with neck dystonia were subject to serious prejudice and discrimination from others in society (Rinnerthaler et al., 2006).

However, there is limited and conflicting evidence as to how stigma affects the wellbeing of people with neck dystonia. For instance, one study found a significant

relationship between stigma and both the mental and physical aspects of quality of life (QoL) in a survey of 289 people with neck dystonia (Ben-Shlomo et al., 2002). However, another study of people with segmental dystonia, affecting two or more parts of the body in areas adjacent or close to each other, found no significant relationship between stigma and health related QoL (Basurović et al., 2012). Both studies were limited because stigma was measured using a questionnaire designed for people with cancer (Macdonald & Anderson, 1984), which had not been validated for use with people with dystonia. Jahanshahi and Marsden (1988) found that negative self-referent cognitions such as self-punitive thoughts and negative body image, which could have been associated with stigma, were the prominent components of depression in torticollis.

The social world is also an important source of support. Social support is defined as the perception or experience of feeling loved, cared for, valued and part of a social network (Wills, 1991). Support can come from family members, friends, work colleagues, religious and other communities, and has been found to contribute to physical health and increased longevity (for example, Berkman & Syme, 1979). Social support can act like a buffer against the negative effects of identity loss following the onset of ill health, and during the period a new identity is constructed (Barker et al., 2014). However, social support has also been found to contribute to unhelpful thought patterns, for example, by reinforcing negative ideas about ill-health or increasing the fear of social isolation (Adams, 2015). An international survey of 1,071 people with neck dystonia found that 36% of respondents reported that when their symptoms were at their worst, family life and relationships were affected (Comella & Bhatia, 2015).

Quantitative studies are limited in that it is difficult to capture individual experiences of how society and identity interact with the physical body. Qualitative studies overcome this limitation by exploring issues in depth. One such study which examined experiences of living with dystonia found that the stigma of an unfamiliar and visible condition had a negative impact on participants' sense of self-identity (Morgan et al., 2021). However, the Morgan et al. study (2021) explored the broad experience of living with dystonia, rather than focusing on specific aspects of that experience, and included many different forms of dystonia, so the experiences of living with specific symptoms were not elucidated. Consequently, the present study aims to build on this evidence by exploring in more depth how people's social interactions are affected by living with a specific type of dystonia. An improved understanding of the experience and impact of neck dystonia will help psychologists and other health professionals when supporting members of this population. The research question was: what are the social experiences of people with neck dystonia?

Method

Design

A qualitative, explorative approach was adopted to understand the topic in greater depth. The study followed an interpretative phenomenological analysis (IPA) approach, as outlined by Smith et al. (2021). IPA is considered a suitable design as it is in-depth and can capture the interaction of social and psychological factors (Eatough & Smith, 2017). The method was originally designed to understand the experience of people with chronic health conditions (Smith, et al., 1999). IPA has its roots in phenomenology, that is, exploration of human experience, and in hermeneutics, that is, how the experience is understood and

interpreted (Smith et al., 1999). The double hermeneutic of IPA relates to the interpretation provided first-hand by the participants, and then second-hand by the researcher (Smith et al., 2009).

Ethical approval for the project was granted by the Lancaster University Faculty of Health and Medicine's Research Ethics Committee (see Ethics section for the full ethics application).

Participants

The aim of an IPA study is to use a small, well-defined, purposive sample (Smith et al., 2009). Individuals were eligible to take part if they (a) were adults between the ages of 35 and 65; (b) self-reported having a diagnosis of idiopathic neck dystonia for at least a year; (c) were English speaking; (d) were able to participate in an interview either online or by telephone. The age range was selected to capture people mid-life who were likely to be (or have been) in employment, in long-term relationships and possibly with caring responsibilities. Using a year since diagnosis was intended to exclude people who were in the initial stages of understanding the condition following a new diagnosis. People were invited to attend regardless of severity of symptoms or whether they were undergoing treatment.

A total of ten participants took part in the study aged 38 to 64 (mean = 54.7 years), all recruited via Dystonia UK, the UK's largest charity for people with dystonia. Seven were female and three were male. All participants were British, and currently living in the UK. Four participants were working, with various employment statuses, and five were retired. To protect

anonymity, each participant was asked to pick a pseudonym for the final report. Demographic details are presented in Table 2 - A.

<Table 2 - A about here>

Procedure

A recruitment poster was shared with Dystonia UK. The charity then shared this poster with its members via its website, Facebook page and newsletter. The poster invited interested individuals to contact the doctoral researcher by email. Potential participants who expressed an interest were then emailed the Participant Information Sheet and Consent Form (see Ethics section). Due to potential physical difficulties associated with neck dystonia, the means of interview were decided by the participants. Five participants chose to take part via MS Teams, and five by telephone interview. Participants were able to ask any additional questions at the start and end of the interview, following which they were sent the Participant Debrief (see Appendix 4 - F).

Data collection

All interviews were conducted by the researcher and ranged in length from 41 to 113 minutes (mean = 63 minutes). A topic guide was developed, informed by previous research relating to social experiences of people with other movement conditions (Barker et al., 2014; Desborough et al., 2020; Theed et al., 2017). Input was sought from the project's field supervisor, a healthcare professional working with people with dystonia, and from Dystonia

UK. An expert by experience was consulted on the overall design on the project and the topic guide.

The topic guide was designed to explore participants' experiences of how living with neck dystonia has affected their social interactions. Questions fell into four broad areas: *Background and clinical information*, including when they were diagnosed, living arrangements, and working life; *Social relationships,* including family, friends and work colleagues; *Context to social relationships*, including being at home, travelling on public transport, hospitals, online world; and *Social identity*, including motivation to socialise and attitudes to other people. Rather than trying to stick rigidly to the topic guide, the researcher took cues from participants as to where they wanted to take the interview. This meant the interview covered topics that were most important to the participant (Smith et al., 2009). See Appendix 4 - A for full topic guide.

Data analysis

Each interview was transcribed verbatim by the researcher. As outlined by Smith et al., (2021) each transcript was analysed in turn to enable an idiographic approach. Step one involved reading and re-reading the transcript to allow for active engagement with the data and to enter "the participant's world" (Smith et al., 2021: p. 80). The next step involved going through the transcript and in a column next to the text, recording anything of interest. By keeping an open mind, the researcher was able to examine semantic content and language on an exploratory level (Smith et al., 2021). The third step involved the construction of experiential statements (Smith et al., 2021). The statements produced are intended to relate directly to the participant's experience and how they are making sense of their experience. The

researcher's interpretation is brought in and combined with the participant's original words, providing preliminary analytic units (see Appendix 2 – B for an example of this process).

The fourth step involved looking for common themes across the experiential statements to develop 'personal experiential themes' (PETs). The fifth step involved naming the PETs and consolidating them in a table. The aim was to capture the essence of the participant's experiences as they related to the research question (see Table 2 – B).

<Table 2 – B about here>

Step six involved repeating steps one to five for each participant so that the researcher had ten tables of PETs for each transcript. The final step was to look for similarities across the PETs in order to create a set of Group Experiential Themes (GETs). This was a dynamic process starting at the level of the PETs but also considering the experiential statements to draw out more of the unique experiences amongst the participants. Researcher interpretation becomes more important during this stage of the process, as inferences are made about how participants' PETs link together. The final table of GETs offer one way of interpreting how participants' social interactions were impacted by living with neck dystonia (see Table 2 - C).

<Table 2 – C about here>

Quality of the data

The principles of Yardley (2000) were followed in order to improve the quality and validity of the research. Yardley emphasises sensitivity to context, commitment and rigour,

transparency and coherence, and impact and importance. For example, all stages of the data collection and analysis process were reviewed by at least one of the two supervisors to improve coherence and impact. As someone who has never experienced a chronic condition that causes pain and exhaustion, the researcher had limited insight into the realities of life with neck dystonia. Therefore, her perceptions were shaped by cultural and societal beliefs and biases related to chronic conditions. This could result in leading questions and the use of certain words that could influence the participants' narratives. However, a reflexive journal was used by the researcher to raise awareness of underlying perspectives and biases that may influence the process (Cresswell, 2012) in addition to discussing these issues in supervision. Further exploration of this is given in the Critical Appraisal section.

Findings

The process of analysis yielded three themes: (1) dismissed by others for having an unfamiliar condition; (2) negotiating a new social identity; and (3) managing the stigma of a visible condition. All participants contributed to each of the themes.

Theme 1: Dismissed by others for having an unfamiliar condition

The first theme related to how participants experienced other people's reactions to them having a rare and poorly understood condition. There was a notable contrast between the distress experienced by the participants following the onset of symptoms and the lack of concern from others in response. This could have been due to other people's unfamiliarity with dystonia or due to a more culturally determined lack of care for people with visible health

conditions. The effect was that participants felt dismissed, invalidated and belittled, leading to ruptures in relationships with family, friends and employers. All participants subsequently spoke of their need to address this general lack of understanding regarding neck dystonia by joining support groups, raising awareness through social media and/or contributing to research.

All ten participants reported the onset of dystonia symptoms as a worrying period marked by the strange, painful and uncomfortable sensations of the head being twisted, pulled and shaken. The path to diagnosis was marked by multiple appointments with different healthcare professionals, often resulting in participants having to return time and again to have their symptoms taken seriously. This seemed particularly hurtful for participants due to an expectation that health professionals should have understood the significance of dystonia. Susan's experience was one of belittling: "*The GP looked at it and said oh you've got a wry neck and the general response of all the medics was that they laughed, they literally laughed.*" Bridget was told by the GP her symptoms were the result of her self-consciousness of being tall, whereas Bridget was "*proud of being tall*".

Once the diagnosis of neck dystonia was confirmed, participants were left reeling in shock at their new reality of living with a chronic condition for which there are treatments, but no cure. Both Aggie and Shadow described how the "*rug was pulled out from under my feet*" (Aggie). Following diagnosis, for all participants, in varying ways, neck dystonia had an all-encompassing and often limiting impact on life. The condition meant participants lost jobs, income, the freedom to drive and use public transport, the ability to enjoy hobbies, exercise and play musical instruments. As Aggie explained: "*I've had to give up work. I can't drive. Everything. I'm uncomfortable or in pain 24 hours a day*". All participants described a feeling

of exhaustion as though they were in constant battle with their bodies: "*I am fighting it all the time, all day, you know, it's, it's always there. So it is really tiring*" (Christian).

Despite the imposition neck dystonia places on the lives of participants, they described being met by others' dismissive attitudes. Rachel experienced such attitudes from her employer: "*I mean she as good as said that she didn't believe the diagnosis*". For Shadow the lack of understanding came from her mother:

"I mean even my mum, we haven't spoken for three or four years now. Because I'm. I'm not the same, you know…She doesn't understand and she's not interested in the fact that I can't do most things.'"

Aggie experienced a lack of awareness from her friends as to how difficult it was to live with physical symptoms of neck dystonia, as she explained:

"You know I've got a few people have said to me, 'oh, yeah I know exactly what you're going through because I clicked my neck two months ago and it was bad'. [But she wanted to respond] *You haven't got the foggiest idea what you're talking about."*

However, there was a recognition by some participants that they would need to be more explicit in order that others could understand. For example, Christian noted how his friends assumed dystonia was similar to a pulled muscle because he had not spelt out how difficult it was:

"The fact that my friends, and it was quite hurtful, never really took the time to understand ... and I think this is more my fault because of the way that I powered through. And I've never said, 'oh God', other than when we're in the pub, 'can you go and get drinks for me?'".

Also, Bridget noted how once she had posted a video online about how hard things had been for her, she received several supportive messages: "*she* [friend] *phoned me up. She said, 'oh I'm crying'. I said, 'why' and she said, 'oh Bridget, I didn't realise'".* This suggests that other people were not all being purposefully ignorant, rather they just needed help to understand. A connection with others built on shared understanding of the condition was an important coping strategy for some participants, as James noted when joining a dystonia support group:

"So immediately there was somebody there I could speak to who had been on exactly the same journey, you know? And it was that was a massive, massive help".

Theme 2 - Negotiating a new social identity

This theme related to how living with neck dystonia led to changes in both the social groups that participants belonged to and the social roles they had participated in. These changes led to the formation of new social identities. The shifting of social groups was easier to navigate for some participants than others. For example, some participants experienced a strengthening of their old friendships with a reduction in the bond of more peripheral friends and acquaintances, whereas others reported a sense of feeling excluded from social groups and this seemed a more distressing experience. Changes in social role, such as not being able to do the same jobs at home or leaving the workplace due to ill health retirement, were also more

difficult to navigate for some participants than others. Such difficulties seemed to depend partly on how much value had been attributed to the participants' pre-diagnosis social roles and how much their self-identity depended on that role.

Given all participants were of working age, the impact dystonia had on ability to work featured prominently in their accounts of living with the condition. For example, Sarah had to stop working due to a head tremor that meant she could no longer focus on computer screens or read for prolonged periods. Work had been an important part of Sarah's identity, acting as a constant thread throughout her life. Having to stop work led to feelings of despondency:

"I have always worked. Apart from when I was a carer looking after my mother… I think I envisaged going on a bit longer than the age I am at work. And now I can't do that. It is upsetting, yeah. Not to have the choice really".

Having to take ill-health retirement from her job meant Shadow lost income and needed to claim state benefits. Work symbolised Shadow's place in the world, it represented her intelligence, her abilities and her contribution to society. Not being able to work meant Shadow belonged to a "*different part of society to the one I was used to being in*". Although Philip found his employer to be supportive following his dystonia diagnosis, he described being moved to a new department to "*gather dust*", reflecting a sense of no longer feeling important or relevant to the operation. Philip experienced a conflict of, on the one hand, needing increased assistance to do his job, and on the other hand feeling patronised by people who tried to help: *"I've got quite a bit of pride I don't like being helped with things, but on the other hand if it helps to get the job done".* Ill health retirement came at a good time for Philip and

allowed him to focus on his hobbies. This showed that leaving work prematurely was not always detrimental to wellbeing.

For other participants, neck dystonia marked the advent of a new way of working. Three participants explained how they now worked remotely. Technology and working from home had allowed them to manage their jobs around physical symptoms, for example, being able to rest when needed, and to avoid customers and clients noticing their symptoms. Managing to overcome difficult periods of work history was described with a sense of pride by some participants. For example, Rachel left her job due to ill-health after her dystonia diagnosis and took the opportunity to start her own business, as she reflected*: "I do wonder that we have to be kind of pushed - don't we sometimes? - to make the change that we want to change."*

Changing work patterns also meant a readjustment of social relationships for some participants. Shadow described how she found herself outside her social "*bubbles*", which had been linked to her employment status: *"They focus on themselves. They're like little bubbles … And once you're out of that bubble, everybody else carries on."* Shadow expressed a sense of disillusionment as she recognised her 'work friends' were not really friends as they did not support her or try to keep in touch: *"I was really only there as a work colleague … And that I find with hindsight a little bit unsettling. Really, I was only there for what I did. I wasn't there for me".* For Shadow, the transition from being able bodied to having a physical disability was like being able to see the world for what it is, like having some truth about the social world revealed.

The physical difficulties experienced by participants also led to lessons being learnt about themselves. For instance, Lucy described a new clarity in understanding of what is important to her: *"I would try to prioritise the people - all the things that I really want to do - and forget the rest".* Some participants reflected on how living with neck dystonia had changed the way they related to other people, for example, becoming more sensitive to others' distress, as Bridget explained: *"I'm nicer, nicer with people. And I think about their, their problems and if they're feeling uncomfortable ... I'd bend over backwards to try and make people feel comfortable."*

Theme 3 - Managing the stigma of a visible condition

The third theme related to stigma including how stigma manifests, how it is experienced, and the lengths to which participants must go to manage its impact. A major feature of neck dystonia is that it is a condition which is visible to others. This had an impact on how participants saw themselves and how they imagined others saw them. Participants repeatedly used the words "*weird*" and "*strange*" to describe their symptoms. For example, Susan described her experience of working in a public facing job: *"They maybe think you look a bit weird. They're thinking, 'why is this weird person talking to me?' You know? 'What's the matter with them?'".*

Several participants reported a sense of being stared at, and even judged or assessed by others:

"If you go shopping or something you notice people's eyes sort of travelling up your head up to your eyes. I know what women must feel like now with men's eyes. It's a very strange feeling" (Philip).

Being looked at by strangers, without being spoken to, left an unnerving gap in the social interaction, which felt one-sided with power weighted towards the stranger without the visible difference. Participants struggled with not knowing what people were thinking about them. Shadow suspected that people were misrepresenting her as someone with less intelligence:

"I think people are seeing that you're not very bright, right? And that's that sounds horrible. And I don't mean it to be horrible. But they talk slower. And they talk louder. There's nothing wrong with my hearing".

James was hesitant to explain what he thought people were thinking about him. He conveyed a sense of being transported back to the playground, where children say cruel, thoughtless things to each other:

*"When I was younger, we used to use the horrible phrase "s******" ...which we probably didn't understand… And so in my mind, that's what I guess I thought people were thinking about me".*

Other people's stares were also internalised by participants and experienced as shame or self-consciousness. Bridget described how she changed from being a confident young person to one who feared being around other people. She blamed this fear on the "*unwelcome friend*" that is neck dystonia. It manifested as a constant voice in her head which commentated on how she looked to other people, exacerbating her self-consciousness: *"I've got this person inside me that even now … it's always there from the moment I wake up to the moment I go to*

sleep". Lucy reported a similar experience of having a split in her mental state: *"I'm trying to have a conversation with somebody, but in the back of my mind I'm thinking about how bad does this look".* Self-consciousness acted as a vicious cycle for some participants, as being stared at made the symptoms worse, which then made the self-consciousness worse, as Christian explained: *"Anxious, anxiety, tiredness, all these different things. It can [exacerbate] the dystonia, so my shaking goes into overdrive, and I feel very self-conscious then".*

The existence of stigmatised attitudes meant Sarah felt blamed for her neck dystonia. This led to her feeling embarrassed to tell people about her condition and even to ask for medical help:

"It feels like always moaning and I feel like that when I contact the doctors too. I know it isn't my fault, but I think you do feel embarrassed and keep on saying there's something wrong. No, I don't really talk about it".

However, Rachel took an alternative position and considered how the visibility of dystonia had the benefit of showing to people that you do need help, and helps other people understand how difficult dystonia is:

"You know, the fact people can see or can't see, it really shouldn't be a big deal. But of course, it is. Because if people can't see that you're struggling with whatever or that you have a condition, then you are not treated any differently".

To manage the impact of stigma, participants described strategies they had developed which had varying degrees of positive and negative consequences. Some strategies were designed to conceal the physical symptoms of neck dystonia, for example, using gestures to

disguise a head tremor, sitting at certain angles to disguise head position and leaning the head on a hand while sitting down. These strategies often came at the expense of causing pain. Some participants reduced the time they spent amongst strangers, thereby increasing the risk of social isolation. Bridget described how she had used alcohol to quieten her self-conscious thoughts, to stop worrying about other people looking at her, and to some extent, to lessen her physical symptoms. This strategy had a significant impact on her daily activity:

"Every single time I went out socially from about 21 onwards, I had to have a drink every time…I feel I don't need to have a drink before I go out, but it's taken all this time and I have, I have made such a fool of myself in family situations".

Mitigating stigma using cognitive strategies was another option. For example, trying to ignore the staring, explaining the condition to people, or accepting that stigma was an inevitable part of "*human nature*" (Christian). Philip used humour to both explain to people that he had a brain operation and to lessen his and others' awkwardness:

"We'd go away for these away-days, and you'd have to introduce yourself with something unusual about yourself – when I used to say I'm part cyborg - that's usually a good ice breaker."

Discussion

This research aimed to understand the ways in which neck dystonia impacts on individuals' social interactions. The themes highlighted how participants experienced the dismissive attitudes of others given the strangeness and unfamiliarity of the condition, how

their social identity evolved following the changes brought on by physical symptoms and how they experienced and coped with stigma. This complexity and challenge in negotiating the social world is interrelated with the physical difficulties, pain and exhaustion that neck dystonia brings.

In the first theme, participants discussed their confusion and shock at the onset of neck dystonia symptoms. This resonates with the findings from the Morgan et al. (2019) paper which described participants as "*struggling to escape the darkness*" (p. 946) as they sought to find answers for their condition. In contrast to the enormity of their own experience participants in the present study were confronted by other people's lack of awareness and understanding. This came from healthcare professionals, employers, colleagues, family, and friends. Words or actions that are intended to exclude and invalidate people has been termed the "disavowal of disability" (Hughes, 2007, p. 681). Hughes (2007) theorises that other people's fears of physical difference and social vulnerability are projected onto those who show such vulnerability. Consequently, a "hierarchy of existence" is created, with people who are non-disabled at the top (Hughes, 2007, p. 681). This can lead to oppression of those at the bottom by those at the top, through psycho-emotional disablism impacting individuals' sense of worth and self-esteem (Thomas, 2007). By being made to feel that their condition was not important, this suggests that participants were experiencing this type of oppression.

The second theme highlighted how a change in social networks and roles impacted on participants' sense of self-identity. Unexpected life changes can change our relationships to other people and how other people see us, which can disrupt our sense of self-continuity (Haslam et al., 2021). The subsequent re-evaluations of social identity matter because they affect the resources people have to cope with such changes (Jetten et al., 2009). Some

participants in the present study seemed to accept the shifting social groups brought on by the onset of neck dystonia, whereas others were more upset and "unsettled" by it. This could relate to the Social Identity Model of Identity Change (SIMIC; Jetten et al., 2009; Jetten & Pachana, 2012), which posits that a person's capacity to successfully negotiate life changes depends on how previously developed social identities provide support to facilitate the establishment of new identities. A meta-synthesis of 16 studies found support from the family while a new identity is constructed can protect against the negative effects of identity loss following diagnosis of multiple sclerosis (Barker et al., 2014). Those participants who found support from family and groups of old friends, unrelated to work, may have been able to more easily adapt to their new identity as someone with a chronic health condition, than those who did not have access to that support.

Changes in social identity can also be related to stigma, as participants reported their discomfort with becoming known as someone who does not work, who is cared for by relatives or needs to claim ill-health benefits. Goffman (1963) describes a stigmatised individual or group as possessing features which differ from a social norm, in this case, the norm of being able-bodied. A large body of research exists which links stigma to visible health conditions (for example, Maffoni et al, 2017; Mayor et al., 2022, Smith et al., 2015). The third theme focused on participants' experience of the internalisation of stigma as shame, embarrassment or self-consciousness. This suggests people endorse negative beliefs and feelings associated with their stigmatised condition and apply them to the self (Link, 1987). Internalised stigma is thought to be more disruptive to an individual's life than enacted stigma, that is, actual discrimination by others (Scambler & Hopkins, 1986). Feelings of self-consciousness had a significant impact on some participants including needing to drink alcohol before social interactions, avoiding social interactions altogether, and avoiding seeking help from healthcare

professionals. Being observed by others led to feelings of uncertainty and misrepresentation, for example, in Shadow's description of thinking people assumed she was less intelligent. This is similar to findings in Parkinson's disease, where the condition was described as a "misrepresentation of the self" (Bramley & Eatough, 2005).

The third theme also showed participants were engaged in numerous strategies to manage and mitigate the effects of stigma. Similar findings were made in studies of people with Tourette's syndrome, who disguise and misattribute their visible symptoms by seeking solitude and disguising tics as intentional movements (Buckser, 2008). Concealment of symptoms by people with chronic health conditions has been found to deter people from seeking social support and medical treatment (Earnshaw & Quinn, 2012). Other strategies participants used to mitigate the feelings of stigma involved cognitive techniques, for example, humour. This is described by Goffman as an act of "covering" and something employed by stigmatised people to help "normal" people feel less uncomfortable (Goffman, 1963). However, there is a risk that using humour can contribute to societal narratives that allow for the condition to be trivialised, as is the case for Tourette's syndrome, and thus, further misunderstood by others (Malli & Forrester-Jones, 2021).

In summary, the findings provide support for the theoretical concept of disablism (Thomas, 2007), in that people with neck dystonia experience both structural oppression and psycho-emotional disablism. Participants were first given the sense from others that their condition is not important, faced structural barriers in terms of work and socialising, and then made to feel that they are "weird" and should conceal their symptoms, leading to feelings of embarrassment. Social support enabled participants to renegotiate their identities as suggested by the SIMIC, and helped to buffer them from the stigma of having a visible, unfamiliar

condition. Morgan et al (2019) found that people with different types of dystonia contend with the stigma of an unfamiliar condition and a perception that dystonia has a psychological explanation. The present study builds on this by exploring how the specific visible and physical features of neck dystonia led to difficulties in social interactions and identity development. Quantitative studies show that psychosocial factors such as stigma and self-esteem have a significant impact on quality of life and mood (Ben-Shlomo et al., 2002), and that illness perceptions are related to distress in people with neck dystonia (O'Connor et al., 2022). Findings from the present study add depth to this evidence by describing the complex ways these interrelated factors are experienced.

Clinical implications

This study has relevance to both clinical psychology and in a broader sense to societal-level systems of power that create and maintain stigma. Given that treatment for neck dystonia is aimed at treating the motor symptoms (Albanese et al., 2019), these findings show that psychosocial features of the condition interact with motor symptoms and contribute to distress. As such, these features should be considered as part of a treatment approach (Van den Dool et al., 2016). The provision of psychoeducation may act as a suitable first-line intervention, as is recommended in the treatment of Tourette's syndrome (Pringsheim et al., 2019), in order to increase positive attitudes to the condition which may in turn reduce distress (O'Connor et al., 2022). Mindfulness-based group interventions have been trialled with people with different forms of dystonia and have yielded positive early results (Sandhu et al., 2016). A three-day group residential programme found participants adopted mindfulness approaches as a coping strategy to challenge unhelpful thinking and the group dynamic legitimated their condition (Sandhu et al., 2016).

Other therapeutic approaches could be considered to help adjust underlying beliefs about an individual's identity and the condition. For example, cognitive-behavioural therapy could be a suitable approach for addressing negative body concept (Lewis et al., 2008) and for helping people to deal with the consequences of self-stigma, such as feelings of embarrassment (Corrigan & Calabrese, 2005). Acceptance and Commitment Therapy has been recommended to help support cognitive flexibility in people with multiple sclerosis, which may act as a buffer between stigma and wellbeing (Valvano et al., 2016). Compassion focused therapy has also been found to be an effective approach to treating shame associated with chronic illness (Carvalho et al., 2022).

The findings also highlight the important role that social identity plays in people's wellbeing. Identity-based interventions could support people to build connections and form new social groups (Schmidt & Ownsworth, 2022) as was identified in the Sandhu et al. (2016) proof-of-concept study. Support groups have been identified as helpful in reducing stigma, as they can provide a forum for sharing experiences, helping people feel validated and reducing isolation (Posen et al., 2001). Delivering psychological interventions, such as those mentioned above, in a group format could also enhance group-based identity which predicts illness-related self-efficacy (Cameron et al., 2018). More research is required to evaluate these therapeutic approaches and their efficacy in relation to people with neck dystonia.

However, it would not be appropriate simply to offer individualised treatment for psychological distress caused by stigma without also addressing its origin. Systemic interventions and structural changes are also required to reduce stigmatising attitudes and provide better support. Hejinders and Van Der Meij (2006) found stigma intervention

programmes should initially focus on an intrapersonal level. This empowers affected individuals to use their expertise to develop and implement programmes at a community level. Effective interventions should include educational campaigns aimed at increasing general public awareness of the condition (for example, Patalay et al. 2017). Given the experience the participants reported during the diagnosis process, educational campaigns should also be targeted at GPs and healthcare professionals. This could include recommendations for health professionals to draw on the resources of people's social relationships (Haslam et al., 2005; Jelinek & Hassed, 2009), for example, by aiming to have a better understanding of their patient's social identities and meaningful activities. The media, academics and charities also have a role in disseminating information, with the help of healthcare professionals, to paint a more accurate picture of conditions that, although life-limiting, can be treated and adapted to. This information should seek to increase the social value of people with neck dystonia by educating others about their continuing role as valued members of society (Reidpath et al., 2005).

Limitations and future research

There are limitations that relate to the design of the study. For instance, the homogenous sample of participants, as is required by IPA, means the results are not generalisable to younger or older people, and those from different cultures. Only nationality was recorded, not ethnicity, which means some British ethnic populations were not represented. Future research involving non-white and non-Western participants would be beneficial. This is especially the case given cultural differences in how stigma is manifested and expressed in relation to chronic health conditions and visible differences (Abdullah & Brown, 2011). Stigma is a social construction which depends on a power differential between

the stigmatised and the stigmatisers (Link & Phelan, 2001). There are other social constructions which create power and privilege in societies, for example, race, gender and age (Rosenthal, 2016). The research is therefore limited by viewing stigma solely through a visible difference perspective. Future research should seek to understand how different social identities intersect when living with neck dystonia.

Although the sample was representative in terms of age and gender for people with neck dystonia, the years since diagnosis varied from one year to 30 years. This could have an impact in terms of adjustment and adaptation to the condition, which could in turn impact how people interact with others. Ben Shlomo et al. (2002) found longer disease duration was associated with better quality of life. The authors suggested this could be related to the development of successful coping strategies over time. Further research could take a qualitative look at how people learn to cope and adjust to the symptoms. Greater understanding of the distress experienced by people with neck dystonia using qualitative approaches could help to develop tailored psychological interventions for this population.

Participants were recruited through Dystonia UK, which could lead to selection bias. For instance, people involved with a charity may be more interested in campaigning because they find such support to be helpful themselves, they may be more likely to be educated people who hunt out such opportunities, or they have experienced more negativity from society than others with neck dystonia. Alternatively, the participants may have been those who were less stigmatised and therefore happy to be interviewed, as very embarrassed and self-conscious people may not come forward to volunteer for research projects. It was notable that all the participants were in a relationship with a partner, husband or wife, and that this relationship was a significant contributor to social support. Further studies could also obtain partner and

family perspectives to understand in more detail the nature of social support and its impact on wellbeing.

Conclusion

This study used IPA to explore ten participants' experiences of navigating the social world with neck dystonia. Participants spoke about the dismissive attitudes they experienced from other people regarding their diagnosis, how their social identities have changed, and how they internalised and coped with the impact of stigma from a chronic and visible condition. These findings could have a beneficial impact on individuals with a diagnosis of neck dystonia as they highlight the important interaction of psychosocial experiences with physical symptoms. Further research to understand the distress experienced by people with neck dystonia would be beneficial in order to tailor clinical interventions.

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Table 2 – A: Participant demographics

Pseudonym	Gender	Age	Years since diagnosis	Employment Status	Relationship status	Children
Shadow	F	55	6	Employed	Married	2
Philip	M	59	13	Retired	Married	2
Susan	F	61	30	Retired	Married	2
Aggie	F	58	7.5	Retired	Married	2
Sarah	F	59	1.1	Unemployed	Married	3
Bridget	F	63	20	Retired	Married	2
James	M	64	14	Retired	Married	2
Rachel	F	46	7	Employed	Partner	0
Lucy	F	38	1	Employed	Married	3
Christian	M	44	15	Employed	Married	2

Table 2 – B: Exploratory statements to personal experiential themes (PETs) and sub-PETs (Aggie)

Exploratory statements	Personal experiential themes	Supporting quotes
Shock of diagnosis – and knowing there is no cure Treatment made it worse Realising treatment would work – lost hope – lost will to live Constant pain and discomfort – that is new life Everything lost – life was over as she knew it Dystonia is a story of loss – nothing is gained Loss of family house was heart-breaking – heart was in the house Having to grieve for lost life and lost future Life now revolves around dystonia	Journey of dystonia – from shock to grief to acceptance • Shock of diagnosis – out of nowhere • Loss of faith in treatment • Loss of current life • Loss of future life • New life of discomfort • Learning to accept	"'I [neurologist said] can't do anything for you at the moment, but you have a brain disease'. Which immediately after that, I didn't really hear a lot because I right, that's it. I'm dead." "My life - that rug was pulled out from under my feet… and became very, very depressed and didn't really want to carry on living after that. But after lots of help and counselling, I've kind of got my life back on track." "I couldn't live the life I was, you know, wanting to live, you know, we met, my husband and I married early. We had our children young, so we thought once they were, you know, married, then we could carry on and have a bit of a life." "I've had to give up Work. I can't drive. Everything. I'm uncomfortable or in pain 24 hours a day. And I've just had to learn to live with it and I'm now." "The whole condition is exhausting. The is no respite from it and all the things you just take for granted. You shouldn't really have to think about your neck. When you're doing something but my constant, you

At different stages of acceptance with husband Had to re-evaluate whole life – don't see it as loss see it as forward step Doesn't want to dwell on negative		know, thing is trying to make myself comfortable all the time. It's physically exhausting and mentally exhausting as well." "Although I'm much more accepting of the situation now, much more accepting. It's still my whole life revolves around this wretched condition."
Tried to go back to work but had to admit defeat – it was over Lost the vision of the grandma she wanted to be – an active hands on one Pain is ephemeral but relationship with grandchildren is real	Renegotiate self-identity after diagnosis • Parts of her identity she has had to give up • Parts of her identity she did not want to give up • Identity/ role of wife/ mother/ grandmother has changed	"Our finances changed because I had to give up work. Eventually, the strain got so bad that my husband took early retirement. He retired at 52 and so on a vastly reduced income. And he's my carer now. We've got two grandchildren and it's really hard because I can't run around and, and play with them." "I've always been, you know, very particular home maker and having to rely on my husband to help me cook the meals, do so many things he has to help. with showering to wash my hair and dry my hair and so all the things that you just so much take for granted. Just vanished."

She doesn't want to be defined by her health complaints – a moaning Minnie Role of husband changed – gone from breadwinner to carer Upsetting to see him in kitchen – that was her domain. Accept she doesn't have a domain – what is her contribution to family now? Identity of homemaker has gone – and has to watch husband take on that role		"Fortunately we have a very, very strong, strong relationship and so it's altered it, but actually we really enjoyed being being together and I I really dislike the personal care aspect of it. Because, you know, you shouldn't have to rely on your husband for that." "[I get] upset that he's [husband] had to take over what I would consider my my domain kitchen." "I still try to be mum, you know, still have them over and try and cobble together a meal and stuff, but they're obviously very upset for the situation that I'm in. But again, I'm very fortunate. They're very kind and loving children." "And is it because those things kind of make up people's identities, their jobs and their cars and their homes and things as your do you feel like your identities changed over that." "I think it's [dystonia] brought out my fighting spirit."
Strong relationship with husband is major support factor Realises how fortunate she is to have a strong support network Relationship with friend has changed as she can't get as involved in social life	Costs and benefits of social relationships have changed • Benefits – there are certain bonds that have strengthened due to dystonia	"They're [cousins] all normal and getting on with their lives and I'm struggling with this [dystonia] and I don't want them to see me like this because I just feel so embarrassed and be I don't want to have to explain it to 21 different people why I'm suddenly like this so. Yeah, one of my cousins remarried 2 years ago and she invited me to evening reception and I was like no, sorry, can't do it. Don't want to expose myself to that, you know."

Dystonia stops new friends forming as avoid situations where that might happen She appreciates friends more – change in balance – she has to give more to them as they are receiving less People around you exert an influence - she can engineer this influence to be a positive one by removing negative people Difference in friends and acquaintances – difference in time known and effort put into friendship People are ignorant of how hard it has been for her	• Costs – there are certain bonds that have weakened because they require too much	"I'm very fortunate that I've got a very, very good set of friends, very close set of friends. So they're all very supportive. I mean, over the last two years we've been doing a lot on zoom and I tend, I tend to keep my camera off. But they've always said, 'oh, you know, but we really like to see you.' I said, 'yeah, I feel so self-conscious.'" "I think you learn who your true friends are, don't you? When you're in a pickle …I can't say if I've lost any friends through this. Umm. I suppose. But probably what's happened is the friends have stayed friends and the acquaintances have disappeared, right? Just say I'm very fortunate. I've got a very, very good set of friends, quite a lot. But the acquaintances are just probably they haven't bothered, and neither have I." "I can't be dealing with people who will always, you know, complaining about their health, who are complaining about their lot in life. And I think, well, you know, I haven't got any choice. I'm. I'm lumbered with this. I'm stuck with it. So I've got to get on with it. If you're complaining, you know I've got a few people have said to me, 'ohh, yeah I know exactly what you're going through because I clicked my neck two months ago and it was bad.' You have got the foggiest idea what you're talking about."

Too embarrassed to leave the house Avoid going out so she doesn't expose herself Expose – reveal something that is happening to her – let everyone see that she has a condition "Abnormality" is emphasised by being around "normal" people People can tell straight away something is wrong with her Having to explain to people means bringing loss to mind Home like safe haven Doesn't want to explain to someone she knows that she has dystonia She would rather them carry on thinking she is "normal"	Mitigating the impact of stigma • Visible condition exposes her truth to strangers • Pretend everything is normal at home	"I'd much rather just never leave the house. Because I just find the whole thing so embarrassing." "Well. I am very very self-conscious of of how I look with the head being constantly twisted and that did have. Well still does have a very big impact on me and my life." "I still don't really go out that much. Mainly because it's uncomfortable. But also because I don't really want to expose myself. To other people." "People are probably not judging you, and you can't tell whether they are or not anyway, what they're looking at. Exactly. But it's still not nice having people. Or even the threat of somebody might look at you." "I I feel I'm safe and I'm secure at home. I know my own surroundings and I can almost pretend that everything is normal when I'm at home. I think it's when I go outside and I'm struggling just to to walk because I can't see where I'm going and properly and I'm worried about what other people are thinking. I think it emphasises to me how abnormal everything is."

Table 2 – C: Contributing personal experiential themes (PETs) to each group experiential theme (GETs)

Participant	**Theme 1 - Dismissed by others**	**Theme 2 - Negotiating a new social identity**	**Theme 3 - Managing the stigma of a visible condition.**
Shadow	Dystonia – strange and unknown A sudden break with the normal	Identity as stories to tell - but not wanting to tell the story of dystonia Work as identity – role in society Income as identity – worth to society Benefits/ needing help from the state as identity Identity from belonging to social bubble People (friends/ colleagues/ family) step up or fall by the wayside	Stigma – into the matrix Society's stigmatised views of disability Stigma of disability lowers people down a social/economic scale
Philip	Regaining control over medical treatment	Loss of role/ identity through needing help	Reclaiming power over what people think about his visible difference

	Becoming active pioneer of medical treatment	Must show to myself I am in control/ I accept my fate	Can't control what others think Others can't help being scared by his looks Using humour to defuse his and others' awkwardness
Susan	Abandoned by medics Learning to cope by herself Power of experience – spreading the word	Dystonia as a separate entity Dystonia can't change her character	Stigma of medical profession Stigma of people staring Mitigating stigma using strategies Mitigating stigma using pragmatism and perspective
Aggie	Parts of her identity she has had to give up Parts of her identity she did not want to give up	Benefits of relationships – there are certain bonds that have strengthened due to dystonia	Visible condition exposes her truth to strangers Pretend everything is normal at home

	Identity/ role of wife/ mother/ grandmother has changed	Costs of relationships – there are certain bonds that have weakened because they require too much	
Sarah	Not wanting to be a moaning Minne Loss of life as she knew it Acceptance as part of life journey/ aging process	End of working life Reduction of social life Loss of retirement future	Embarrassed that she caused dystonia Embarrassed about having to explain dystonia – draw attention to it Embarrassed about seeking help
Bridget	Dystonia has all-encompassing limiting impact on life Family narrative of hiding it/ not talking Doctors telling her its her fault Finding an outlet	Conflict of identity - Outgoing vs afraid of people Dystonia has changed her	Dystonia as the constant commentator Alcohol to quieten the voice Discrimination from others Stigma makes the shaking worse Stigma for having something “wrong” with her

			Strategies to minimise stigma
James	He was dismissed by medics He dismisses his own experience	Feeling validated by dystonia community Separation of dystonia and self – conflict Support – limits on social circle Needing to rely on close circle so he could focus on symptoms	Self-consciousness as uncertainty He knows people look at him Tell people what's wrong to remove the uncertainty – be robust Take control through education
Rachel	Power balance with doctor and patient Feeling dismissed by medical profession She's a scientist – she's just as important as dystonia doctors	Outsider as don't want to be defined by dystonia Practical support means she can carry on life as normal Some people step up some fall into background	Visibility means she is believed – disclosure First thing to support is to be believed

	Insider or outsider of dystonia community? Insider helps perspective, knowledge, advocacy		
Lucy	Don't want to be a 'bother' to people Unfamiliarity of dystonia to others is barrier to other's understanding	Not giving in to the pain – battle between herself and dystonia Reduction in socialising Prioritisation of people she spends time with Consolidation of introvert tendencies	Dystonia means a life of self-consciousness. Strategies to conceal symptoms from others Avoiding social interaction to avoid being the focus
Christian	Lack of awareness in healthcare profession Exhaustion of constant symptoms – hard for anyone else to understand	Dystonia identity combined with new identity of becoming a dad Finding a job to fit round symptoms Support from family (using humour)	Feeling self-conscious (especially of potentially losing balance) Vicious cycle - anxiety makes symptoms worse Stigma from thinking dystonia is a psychological condition

	Biggest struggle with friends – lack of acknowledgement/ understanding/ effort		Strategies to hide symptoms

Appendix 2 – A: Additional quotes to evidence findings

Participant	Theme 1 - Dismissed by others	Theme 2 - Negotiating a new social identity	Theme 3 - Managing the stigma of a visible condition.
Shadow	"I mean even my mum, we haven't spoken for three or four years now. Because I'm. I'm not the same, you know, and I'm, I'm. So even though you know she's family, it's not. She doesn't understand and she's not interested in the fact that I can't do most things. She just wants to carry on, you know, it's fine. I'll just go, 'OK, go ahead. I can't do it'. So there's been no contact there for years."	"I was really only there as a work colleague … And that I find with hindsight a little bit unsettling. Really, I was only there for what I did. I wasn't there for me". "And it just, it was just a disaster, you know, because financially and all the rest of it. I have no money. Where was it? No income, you know, nothing. And that kind of puts you in a different. You're in a different part of society to the one I was used to being in."	"I think people are seeing that you're not very bright, right? And that's that sounds horrible. And I don't mean it to be horrible. But they talk slower. And they talk louder. There's nothing wrong with my hearing". "And it made, it's made me think … when I was normal, I suppose, for want of a better word. Did I do the same thing? If there were people in wheelchairs and things, do you do the same thing to them? Without even thinking about it. And I was put into that world that I thought. **'**Yeah, people do really speak to disabled people like this'".

Philip	"Back in the early 2000s the detail wasn't too good so I didn't have a lot to refer to. I had a few printed leaflets off the neurologist but yes, not a lot. I found the Dystonia Society which was quite helpful. And luckily they were just down the road near my office, the NCA, which is on the Embankment. I would just trundle down there at lunchtime when if I needed if I needed to speak to people if I needed help."	"And I got moved to a department where I think I was basically dumped until I got to my retirement age which I was going at 60 which would have been this year. So they just put me in this place to run, gather dust and I didn't really like it and I didn't understand what was going on no one had time to explain it." "I've got quite a bit of pride I don't like being helped with things, but on the other hand if it helps to get the job done". "It's changed my relationship with my wife … sometimes I think she comes home from work and the last thing she feels like doing is look after me".	"Er it depends on what sort of mood you're in. It can feel like you're a freak. If you go shopping or something you notice people's eyes sort of travelling up your head up to your eyes. I know what women must feel like now with men's eyes. It's a very strange feeling." "I just ignore it to be honest. There's nothing I can do about it. If I'm going to be with them for a long time, I explain to them what it is and that often helps". "We'd go away for these away-days, and you'd have to introduce yourself with something unusual about yourself – when I used to say I'm part cyborg - that's usually a good ice breaker."
Aggie	"[Neurologist said] 'I can't do anything for you at the moment, but you have a brain disease.' I didn't really hear a lot	"I think you learn who your true friends are, don't you? When you're in a pickle …I can't say if I've lost any friends through this. Umm. I suppose. But	"I feel I'm safe and I'm secure at home. I know my own surroundings and I can almost pretend that everything is normal when I'm at home. I think it's when I go

	because I was right, 'that's it, I'm dead'". "You know I've got a few people have said to me, 'oh, yeah I know exactly what you're going through because I clicked my neck two months ago and it was bad'. [But she wanted to respond] You haven't got the foggiest idea what you're talking about."	probably what's happened is the friends have stayed friends and the acquaintances have disappeared, right? Just say I'm very fortunate. I've got a very, very good set of friends, quite a lot. But the acquaintances are just probably they haven't bothered, and neither have I". "I've always been, you know, a very particular home maker and having to rely on my husband to help me cook the meals … help with showering to wash my hair and dry my hair. that you just so much take for granted. Just vanished."	outside and I'm struggling just to, to walk because I can't see where I'm going properly and I'm worried about what other people are thinking. I think it emphasises to me how abnormal everything is".
Bridget	"You know I get a sore neck that I have Botox for and um but that's just the surface but when you start digging deep to the day it began." "My dad died when I was eleven, about four years before that and I it was nothing to do with that whatsoever.	"I'm nicer, nicer with people. And I think about their, their problems and if they're feeling uncomfortable … I'd bend over backwards to try and make people feel comfortable."	"I've got this person inside me that even now … it's always there from the moment I wake up to the moment I go to sleep". "Every single time I went out socially from about 21 onwards, I had to have a drink every time…I feel I don't need to

	But they [school nurse and teacher] were thinking it was that, you know, and, um, so I put up with it for a while and I didn't tell my mom because my mum was busy."		have a drink before I go out, but it's taken all this time and I have, I have made such a fool of myself in family situations".
Susan	"The GP looked at it and said oh you've got a wry neck and the general response of all the medics was that they laughed, they literally laughed."	"So I found that this gave me a real empathy for people with physical things like cerebral palsy and spasticity because I could understand to a small extent how physically draining and whatever so it helped me to support them in a manner that I hope worked for them well. I think he did, because, you know, they always seem to like me."	"They maybe think you look a bit weird. They're thinking, 'why is this weird person talking to me?' You know? 'What's the matter with them?'". "So they probably just looked at me and thought, 'oh she's a bit strange looking' and for me, but because I'm naturally upbeat and smiley. And because I'm pragmatic you know you've just got to get on with it".
Sarah	"So people don't know what it is and I've never heard of it. And when you say cervical dystonia, I think they think it's women's problems…It's embarrassing."	"I have always worked. Apart from when I was a carer looking after my mother… I think I envisaged going on a bit longer than the age I am at work.	"It feels like always moaning and I feel like that when I contact the doctors too. I know it isn't my fault, but I think you do feel embarrassed and keep on saying

		And now I can't do that. It is upsetting, yeah. Not to have the choice really". It's just everything if I have visitors in, we have people in for coffee last week and it's looking from one person to another, it sets it off. I Start to feel really quesy. I've had enough after now, it just puts me off the socialising."	there's something wrong. No, I don't really talk about it".
James	"Felt a bit of a fraud going there when you see people who had brain injuries and car accidents and things and I went there with a slightly bent head, but they [hospital] were superb."	"So the work were very, very supportive of me, which again. Was, you know, a fantastic thing because I didn't feel under any pressure. I was able to sort of concentrate on getting recognition, you know, managing the symptoms, if you like, without any sort of threat to my job security, if you like." "Stumbling, you know, as the payments went up and down, or if you were, you know, on the edge of the pavement that you might just miss it. You know … it was a physical, physical problem,	"[I] would get odd looks from people as you walk along the street. You know in your head... You don't know whether you are perhaps oversensitive yourself, but you know you would get strange looks from people sometimes". "The fact that you look a complete weirdo when your head is, you know, craning in these strange directions… I wonder what they're thinking of me, you know, are they thinking of, you know, what are they thinking about me?".

		walking along the road at times. And it actually, it did concern me because I've always enjoyed physical exercise and walking was one of big the things I did."	"When I was younger, we used to use the horrible phrase "s******"…which we probably didn't understand… And so in my mind, that's what I guess I thought people were thinking about me".
Rachel	"they [doctors] just sort of said 'oh it's probably just stress' and anyway they didn't give me anything for it and it was a bit of a waste of time." "I mean she [employer] as good as said that she didn't believe the diagnosis".	"You know, the fact people can see or can't see, it really shouldn't be a big deal. But of course it is. Because if people can't see that you're struggling with whatever or that you have a condition, then you are not treated any differently, you know? And, and it's not about needing to be treated differently. It's about needing to be seen, I think, for people, isn't it?".	"It's not something that any of my online clients know anything about or my my private clients now know about because they just never needed to. It would only have been something I would be discussed if I were symptomatic. But because it's so obvious, you don't really have a choice in that regard, you know? So, I mean, it's like, yeah, you're gonna look really weird… It does look weird, right?"
Lucy	"I haven't really talked to [husband's] side of the family. Just seems to be like	"I would try to prioritise the people all the things that I really want to do and forget the rest".	"I'm trying to have conversation with somebody, but in the back of my mind

	it's a big thing for me, but it's not really that big a deal… it sounds like a bit of a whinge if I bring up 'ohh, by the way'. It's not like an illness. You know what I mean? It's not like I broke my arm or I've got cancer. It's just the thing I've got."	"I'm not prepared to live a life of doing nothing, so I might as well carry on doing stuff. It's gonna hurt whether I do stuff or not." "I've got three kids, so your social life is never that brilliant is it when you've got kids? But definitely do that less now [go out to restaurants]. You're thinking about…how much discomfort I'm gonna be in, sat in the same bit for ages."	I'm thinking about how bad does this look" "I'm really conscious of…I have to do like a Teams meeting, all I can see my little head in the corner bobbling, you … any situation where I'm sat directly opposite somebody very conscious about it, because I know I can't stop it." "I'm conscious of people noticing it, and the added bonus of to sit in that position for a long time makes it worse. When I'm going about my day, I've kind of learned to disguise it a bit so I'm never still, try not to be still."
Christian	"I actually had to tell my doctor that I had cervical dystonia. And at the time my doctor said, 'no, you haven't'". "The fact that my friends, and it was quite hurtful, never really took the time to understand. I I think for them, and I		"Anxious, anxiety, tiredness, all these different things. It can [exacerbate] the dystonia, so my shaking goes into overdrive, and I feel very self-conscious then". "Unfortunately, it's human nature that people will look"

	think this is more my fault because of the way that I powered through. And I've never really, you know, said, ‘oh God’, other than when we're in the pub, ‘can you go and get drinks for me?’. I've never really spoken too much to them about it because they for them I think it's the same as having a pulled muscle or something like that”.		

Appendix 2 – B: Transcript extract: Initial coding of exploratory notes and experiential statements (Shadow)

Experiential statements	Transcript	Exploratory notes
Diagnosis provokes stigma in others Other people reflect her own fears about the impact of her condition Other people project their own fears of ill health on her Experience of stigma through the gaze of others – the way she is looked at A major fear is having to tell other people about the diagnosis	And I said this, and he looked at me and said "Oh s***". And I thought that's really not what I needed right this moment 'cos I think it was at that point when I thought this is not good, this is not good news, you know. And it was just the way he looked at me and said nothing apart from that. And I thought…yeah, this is not good news. Um, but since that initial diagnosis, it was at least six months before I saw a consultant, by which time the doctor parent had signed me	Other person's reaction to her diagnosis was negative Negative, reductionist, final statement She has a clear memory of this instance, which happened over 6 years ago – it must have had a significant impact He affirms her fears Or did he himself look afraid? His own fears of illness "the way he looked at me" – feels derogatory and offensive "news" – her diagnosis is something she has to announce to others Negative expectation about having to tell employer

Difficulty in understanding/ accepting that dystonia is incurable	off sick for the rest of that year, so 2016, he said "you'd there's no way you'll be ready to do anything before 2017". And of course then as it went on you kind of sort [know] of the consultant and and I think it was his name was [Consultant] and he sat there. October time 2016, maybe, and he said "It's incurable, you know that, don't you?".	Wait to see consultant Signed off work by GP Told by the doctor she wouldn't be able to do "anything" – sounds very disruptive, all-encompassing Told quite bluntly by consultant that there is no cure Or is consultant sounding patronising Feeling talked down to by the consultant?
Shock of being told her future plans are lost – told in a flippant, dismissive, condescending way by consultant	And I thought, yeah, I'd kind of read up a bit by this. Um, and he said "it's, it's not going to go away." No I kind of guessed that.	She is emphasising the finality of dystonia – the huge shift from pre-dystonia to post-dystonia Consultant explaining what "incurable" is. In a condescending way or trying to emphasise seriousness?

	"The best you can hope for is tolerable management." Well, what exactly does that mean? He said "well, how much can you cope with? Is how tolerable it is for you".	The doctor described it as a relative thing – suggesting she could continue with previous activities if she can cope with the pain of dystonia? That doesn't sound acceptable to her.
Major concern is how diagnosis will impact career Lack of empathy from consultant about how diagnosis will impact career – conversation could have been more sensitive and constructive Shock of thinking life as she knew it was over	And I thought, yeah, this is the end of my career and and that's what I said to him. I said I'm not going to be able to do this teaching and playing. And he's "well, realistically probably not" and and it was then you know so six months down the line it was kind of, yeah, this is the end of what I know. And it's just that the rug was just pulled. It was just a whole different ball game at that stage.	Thoughts are around how prognosis impacts career 6 months on from first symptoms she feels like her world has ended Expression of rug being pulled to explain quickness and all-encompassing change Idiom of ball game – new rules, new scoring system, new team, different winners and losers

Going from a period of safe certainty to unsafe uncertainty	Of right what? What do I do now? And I'm sure you'll hear that from a lot of the people that that you're talking to. It's a very sudden.	Feeling of uncertainty Asking others for advice Suddenness of symptoms, but delay in diagnosis, are common themes among people with dystonia
Shared experiences of people with dystonia – confusion, delays, shock but confronted by flippant response of doctor Something she to learn to accept	It kind of takes a while for people to say, well this is what you've got, this is what you've got, you know and you think, ohh yeah, great, but then this is what you've got. And it will be forever. Takes a bit of a longer time to get to grips with and I'm not, I'm not sure entirely I've got to grips with it yet still.	Incurability is something that needs to be "gripped". Idiom of gripping something. Getting hold of so you can steer it/ so it can't escape? Concept of acceptance – this is something she can accept but she's not accepting it yet

Appendix 2 – C: Journal of Health Psychology: Instructions for authors

Manuscript Submission Guidelines: Journal of Health Psychology

This Journal is a member of the Committee on Publication Ethics

Please read the guidelines below then visit the Journal's submission site http://mc.manuscriptcentral.com/jhealthpsychology to upload your manuscript.

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Only manuscripts of sufficient quality that meet the aims and scope of Journal of Health Psychology will be reviewed.

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8. What do we publish?
   1.1 Aims & Scope
   1.2 Article types
   1.3 Writing your paper
9. Editorial policies
   2.1 Peer review policy

1. What do we publish?

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(d) Open peer commentaries on recent articles in this journal or topical issues (up to 2,000 words counting 500 words per table and figure).

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2. report all pre-registered analyses in the text, or, if there were changes in the analysis plan following preregistration, those changes must be disclosed with explanation for the changes clearly distinguish in text analyses that were preregistered from those that were not, such as having separate sections in the results for confirmatory and exploratory analyses (these changes are added as a separate document linked to the text of the main paper)

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We recommend that authors consider looking at the below guidance:

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- Words Matter

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6. On acceptance and publication

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Section Three: Critical Appraisal

Word Count – 3,688

(excluding title page and references)

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Prepared for: Journal of Health Psychology

Critical Appraisal

The aim of the critical appraisal is to allow for further reflections on the project's findings and on the research process. I will first summarise the findings from both the literature review and the research paper, considering the links between the two and implications for clinical practice. I will then consider the strengths and weaknesses of the methodology used and potential future research projects which follow on from this. Personal reflections will follow, focusing on why I chose to study this topic and the role of clinical psychology in furthering research and practice in this area.

Overview of findings

A systematic review and meta-ethnography of qualitative papers related to coping with Tourette's syndrome (TS) was conducted. 16 papers were included, 12 looked at individual perspectives and four focused on parent perspectives. Of the 12 individual perspective studies, six focused on adults and six focused on children and adolescents. Three main themes relating to how individuals and families cope with TS were constructed: (1) redefining the self and social identity; (2) controlling the body; (3) challenging the narrative. Findings were considered in relation to existing literature and implications for clinical psychological practice were drawn out.

The qualitative research study described how people with neck dystonia experience social interactions. An interpretative phenomenological approach was taken (Larkin et al., 2022). Ten semi-structured interviews were completed and transcribed verbatim. Analysis generated three themes: (1) Dismissed by others for having an unfamiliar condition; (2)

Negotiating a new social identity; (3) Managing the stigma of a visible condition. These findings were discussed relative to existing literature and consideration was given to how psychological support could address the difficulties identified.

Similarities and differences

Despite the difference between the two papers in terms of clinical populations and the focus being on different aspects of living with these conditions, there were commonalities between the findings. Both papers highlight how important the social component is of a biopsychosocial approach to understanding health conditions. The theme of social identity was raised in both papers and could be related to the Social Identity Model of Identity Change (SIMIC; Jetten et al., 2009). This model describes how life transitions involving identity changes can be positively adapted to when people are able to maintain pre-existing social group memberships or to develop new ones. In the research paper, participants reported how neck dystonia led to strengthening of existing social groups and relationships, and the weakening of others, thus highlighting how sources of support are affected by life transitions (Jetten et al., 2009). This was unsettling for some whose membership of certain social groups held important meaning and purpose and therefore, leaving these groups had a distressing impact on self-identity. For example, for Shadow, her membership of professional groups prior to her diagnosis also gave her social status and signalled her competence and intelligence to others. Having to leave these groups due to ill-health led to a difficult emotional disruption. The literature review showed how participants with TS could accept their new identity as someone with TS through being accepted and supported by friends, parents, partners and teachers. Relating this to the SIMIC suggests that social support provided continuity for people as they renegotiated their identities. This could be particularly important for adolescents given that

this is a time of transition to adulthood (Kroger, 2006), and could impact on how adults with TS are able to cope with the condition.

Stigma was also identified as a key aspect of the experience for both people with neck dystonia and people with TS and their families (Gofman, 1963). Both conditions are visible and suffer from a lack of public understanding (Gharzai et al., 2020; Fat et al, 2012). For participants with TS, especially the families of those people, correcting misinterpretations about the condition was a key feature of coping (Ludlow et al., 2018; O'Hare et al., 2006; Pine et al., 2022, Travis & Juarez-Paz, 2006). These misinterpretations were namely that others thought that people with TS had some control over their symptoms and that those symptoms would involve swearing (Edwards et al., 2017; Malli & Forrester-Jones, 2021). For participants with neck dystonia, the focus was more on a lack of knowledge from others and the dismissive attitudes encountered from friends, family and healthcare professionals. This could link to the theory of psycho-emotional disablism (Thomas, 2007; Reeve, 2011). This refers to how psycho-emotional wellbeing, such as self-esteem and self-confidence, is undermined by negative social interactions (Reeve, 2011). Both the TS and neck dystonia participants sought to redress this form of oppression by educating others about their conditions.

Mitigating the effects of stigma was important for both TS and neck dystonia groups, and in large part this meant concealing symptoms from others. People with neck dystonia referred to their symptoms as constant battle, that they were always trying to fight against the pulling and twisting of their heads. They were sometimes able to conceal their symptoms by sitting at certain angles to other people, but they were not able to stop their symptoms. TS was also described as a constant, irritating presence (Smith et al., 2016). However, there was an

element of being able to control the tics and to supress them for a short time (Lee et al., 2016; 2019; Wadman et al., 2013). This could explain why the theme of control came out more strongly in the TS sample than the neck dystonia sample. Suppression of tics had the negative effect of increasing the stigma associated with TS as stigma is increased when people are perceived to be responsible for their health condition (Buckser, 2008). Linked to this, Morgan et al. (2009) found that people with dystonia feared psychological explanations for their condition, for example, being told by others that it was "all in your head". This had the implication that they were responsible for the condition and "unworthy" of treatment (Morgan et al., 2009). Although fear of psychological explanation did not emerge in the findings of the present empirical paper, there was a sense that the seriousness of neck dystonia was not believed or acknowledged by others.

Clinical implications and future research

Recommendations for clinical practice emerge from both papers. Psychological therapies can help with identity issues and with the feelings of shame associated with internalised stigma. There is evidence that compassion-focused therapy and cognitive-behaviour therapy are particularly suited to these needs (Corrigan & Calabrese, 2005; Leaviss & Uttley, 2015). Acceptance and commitment therapy (ACT: Hayes et al., 1999) has been found to reduce distress in people with Parkinson's disease through techniques to increase psychological flexibility (Hill et al., 2017). However, any therapeutic approach designed for people experiencing neurological conditions would need to consider specific physical and neuropsychological difficulties. For example, head tremors and pain associated with neck dystonia that make focusing on people and screens difficult (Defazio et al., 2013), and attention difficulties often associated with TS (Khalifa, 2006). Future research would be beneficial to

deepen understanding of distress in these two populations with the aim of creating targeted therapeutic approaches. Research which explored the experience of family members of people with neck dystonia would be useful to understand the social dynamics. Similarly, further research could seek to understand the experience of adults with TS and their partners.

However, it is not just the responsibility of people who are stigmatised to deal with the impacts of stigma and there is a requirement for society-level stigma management interventions (Corrigan, 2005; Heijnders & Van Der Meij, 2006). Given the similarities in the existence and impact of stigma across the two clinical populations, it would suggest that generic stigma reduction interventions could be useful. Cross et al. (2011) developed a Stigma Intervention Matrix, to provide a framework for interventions that can be adapted to specific health conditions and circumstances. The matrix addresses the different components of stigma - labelling, stereotyping, separation, status loss, discrimination – at three different levels – intrapersonal, interpersonal and organisational/ institutional (Cross et al., 2011). Future research could consider how this matrix could be applied to the development of specific TS and neck dystonia stigma interventions, or a more general visible neurological condition stigma intervention.

Reflections on the IPA methodology

Various decisions were made when deciding how to answer the research question for the empirical paper, that was: what are the social experiences of people with neck dystonia? The decision to use IPA was justified because little evidence exists on the experience of people with neck dystonia. Qualitative research enables the researcher to understand the participant's experience of a particular health condition within their own specific social reality (Bryman et

al., 1988). IPA explores how people ascribe meaning to their interaction with the environment which means it is especially suited to studies that relate findings to biopsychosocial theories (Biggerstaff & Thompson, 2008). However, there are also limitations in the epistemology of the approach. For instance, individuals are limited in their ability to express what they experience due to the limits of language (Gough, 2016). Also, by introducing another person, the researcher, to analyse the experience adds to the risk of interpretation bias (Armstrong et al., 1997). To avoid the risk of misinterpretation it is important when using IPA to acknowledge the role of researcher and explore it through reflexivity (Biggerstaff, 2012; Gough & Madill, 2012). By doing this, researcher subjectivity can be seen as a useful resource (Gough; 2016). I aimed to enhance this through supervision and the use of a reflective journal (see personal reflection section).

A small, purposive, homogenous sample is required for IPA (Smith & Osborn, 2008). Although the participants were similar on required factors of health condition and age, they differed in terms of time since diagnosis. I had specified that participants should have at least one year since diagnosis. However, this resulted in a wide variation, from one year to 40 years. This could have been a factor in the divergence within all the themes identified. For example, in the theme regarding dismissive attitudes from others, the participant who had reported the strongest concern for being dismissed by health professionals – the participant who was laughed at – is also the participant who was diagnosed the longest time ago. Although not wanting to diminish her experience, it could be that there was less knowledge about neck dystonia at that time which has now improved. In the theme relating to negotiating social identity, divergence could relate to years since diagnosis as it takes time to develop a new identity and find new social groups (Deaux, 1991). In the theme relating to stigma, those with the longest time since diagnosis may have found ways to mitigate the stigma so do not

recognise it as readily as they might have done when newly diagnosed (Link & Phelan, 2013). However, participants in any sample could never be truly homogenous and there are many ways people differ. The key issue as to how homogenous the sample is should relate to the research question (Murray & Wilde, 2008). Future research could investigate how time since diagnosis impacts social interactions using a quantitative cross-sectional design.

There are no definitive rules as to how big a sample size in an IPA study should be, but smaller samples allow for deeper understanding (Smith, 2011). The sample size in my research paper of ten participants allowed for a rich and broad analysis. However, given how rich the individual participants' interviews were, I could have potentially used a smaller sample which would have given more space for each participant in the analysis (Sandelowski, 1995). Participants were recruited through Dystonia UK's newsletter and social media. Given this is a charity with a variety of roles for its members, including supportive groups, research, fund raising and political advocacy, there may have been a bias towards participants who were interested in these aims and therefore more likely to talk about societal and community issues such as stigma.

The topic guide was created by looking at existing research on the topic (as advised in Murray & Wilde, 2008) but also from speaking to an expert by experience, research supervisor and field supervisor, who is a clinical psychologist with experience working with people with neck dystonia. This enabled me to tailor questions that would be more relevant to people. For example, asking the question about restaurants as a feature of social life was included following advice that people with neck dystonia can find it difficult to eat. However, my first question was to ask participants about their diagnosis, and I referred to this as "from the beginning". This may have drawn people into a narrative trap that they are telling a story that has a

beginning, middle and end. It is a culturally universal feature of human social development that we construct stories (Fisher, 1984; Sugiyama, 1996). Narrating a story with a temporal ordering of events helps people to make sense of their experiences (Singer & Bluck, 2001). This may be because stories allow people to integrate cognitions and emotions (Smyth et al., 2001). However, people may have felt pressured to fall into a socially acceptable story of transformation when confronted by ill health (Miczo, 2003). A typical story of this type which starts with a difficult beginning (the diagnosis) could pressurise participants to end the story with a message of how they overcame the difficulties and emerged as a new and "better" person. Given that one of the themes involved identity, in hindsight it would have been interesting to ask people how they interacted with people before the diagnosis to start the story at a different point in time to fully address the transformation of self and avoid the risk of encouraging participants to fall into a typical story arc. The differences between pre and post illness identity could be another interesting avenue of research.

Several participants explained how although they couldn't socialise and work like they used to, they had learnt to think positively in the face of ill health, which could have represented another socially acceptable story (Bergen & Labonté, 2020). The pressure to rely on socially acceptable interpretations of ill health could be part down to the nature of an interview with an academic researcher. There is a power imbalance inherent in this situation (Miczo, 2003). The researcher could be perceived to be privileged in terms of health and employment status. The participants therefore could be pressured to create a positive identity to make up for a perceived relative lack of power (Miczo, 2003), or to use the interview as an opportunity to regain lost power by acting as a storyteller (Riessman, 1990). One participant said on a few occasions that they did not think they were interesting enough to be being interviewed and questioned

why they had volunteered. This could have reflected someone's reaction to the power imbalance and the pressure to take centre stage as the storyteller.

Such factors as power dynamics can interfere with the collection of a true, authentic account of the participant's experience (Miczo, 2003). However, believing that an interview can achieve such authenticity may be naïve (Sandelowski, 2002). Interviews are social constructions and accounts may be part of a narrative strategy employed by the participant to communicate something other than the truth, for example, aggrievance at poor medical treatment (Sandelowski, 2002). An important way of mitigating against such factors when using the IPA method is to highlight both convergence and divergence between participant accounts (Nizza et al., 2021). Future research could look to integrate different IPA studies exploring lives with chronic health conditions to understand in more detail the divergences in narrative arc.

Personal reflections

Although I have no personal experience of dystonia, I became interested in the topic when looking at the Dystonia UK website and reading the personal biographies of people with the condition. The stories of how the onset of dystonia symptoms were sudden and had significant effects on people's lives had a powerful impact on me. I tried to envisage how I would have coped with this condition and how difficult it would be to manage alongside family and work commitments. Additional background reading and discussions with my supervisors allowed me to focus on areas of interest and specify a research question.

I was surprised when conducting the interviews as to how emotionally salient some of the interviews were. Some participants felt relieved to be able to tell their story of how hard it has been to live with neck dystonia and how grateful that my research represented professional interest in the condition. The emotional nature of some of the interviews led to me experiencing a conflict of role. On the one hand I was a researcher seeking information, and on the other hand, a trainee clinical psychologist who had training in specific skills that could have been helpful to relieve distress. There is an ethical dilemma in whether to use those distress relieving skills when this has not been contracted for by the participant (Allmark et al., 2009). I found some elements of the interviews sad to hear and this led to a determination to "do justice" to the participants' stories. Having to interpret participants' words into personal experiential themes and then consolidate these into group experiential themes, I worried that I had lost the depth and content along with some of the detail. I experienced anxiety of not wanting to speak on behalf of participants and this led to some reticence to include my interpretations in the findings. This conflict has been recognised by Larkin et al. (2021) who emphasised that the double hermeneutic of IPA means that emerging themes are a product of collaboration between researcher and participant. Access to regular support and supervision has been essential to think through these issues. These conversations were also recorded in my reflective journal, an extract of which is as follows:

The idea of belonging to different social bubbles has made me think about which ones I belong to. I can hear from [participant's] story how she felt rejected by her work colleagues and shocked at how superficial those 'friends' were. She had to experience ill health for the superficial relationships to crumble away and be left with the truth about people. It's reminding me of The Matrix [the film] of how once you see the true nature of people and how society is really structured, you can't then unsee it.

Supervision also allowed me to identify my own biases and assumptions so it was easier to suspend these whilst focusing on what was presented in the data. This involves the concept of 'bracketing' (see Fischer, 2009) and the suspension of critical judgement and engagement while analysing the data (Spinelli, 2002). In light of prior research interests, for me the biggest challenge was to avoid looking at evidence with a political lens. The political nature of stigma and social identities were one of the reasons I wanted to study visible conditions. However, it was important not to approach the research question with a specific stigma lens and assume participants would be contending with stigma. Instead, this theme was created from the evidence. Supervision was also very useful for generating ideas as to how emergent themes could be interpreted and to enhance consistency and coherence of the analysis (Yardley, 2008).

Role of clinical psychology in society

This study highlights the importance of the social element of living with neurological health conditions. Both the research paper and the literature review highlighted how stigma affects people and in the case of children with TS, their families. Clinical psychologists have an important role to play in addressing stigma in society as researchers and advocates (Earnshaw, 2020). As researchers, members of the profession can continue clarifying the experiences and impacts of stigma among people with different health conditions. As advocates, psychologists can call for changes in policies that promote understanding in healthcare and community institutions, workplaces and schools, for example, encouraging the use of stigma-free language.

The stigma of health conditions intersects with other systems of oppression such as race, age and gender discrimination (Rosenthal, 2016). These systems of marginalisation and privilege are interwoven and reinforcing (Kerr et al., 2022) and addressing them would require broad, structural-level changes that promote equity and social justice. Rosenthal (2016) recommends that psychologists can contribute to this through (a) engaging and collaborating with communities, (b) addressing and critiquing societal structures, (c) working together to build coalitions, (d) attending to resistance in addition to resilience, and (e) teaching social justice curricula.

For instance, Pachankis (2018) has called for evidence-based mental health treatments for sexual and gender minority populations that are tailored to address life experiences which are not shared by heterosexual and cisgender individuals, including those related to stigma. The present study suggests it is also important to call for similar psychological treatments for people with health conditions which address the specific social challenges of living with these conditions and attend to individuals' intersecting social identities that create privilege and oppression.

Conclusion

This thesis explores the impact of neck dystonia on people's social interactions and how individuals and families cope with Tourette's syndrome. Both neurological health conditions share the characteristic of being visible to others. The research found that the dismissive attitudes of others and misinterpretations have a negative impact on people with the two conditions. Living with the conditions had an impact on identity and for people with TS, the research found identity management was an important part of coping with the condition.

Given the presence of stigma towards visible health conditions, both sets of participants felt they had to disguise or supress their symptoms.

There are strengths and weaknesses of both papers, and this helps to inform future research. Greater understanding of the distress caused by social pressures can inform clinical interventions to help improve the wellbeing of people from these populations. Of course, it cannot be the sole responsibility of individuals to tackle stigma, especially those that have been disempowered by a stigmatised condition. Instead, the profession of clinical psychology should continue to contribute to breaking down stigma through research and advocacy. This includes taking account of the intersectional identities which create oppression and lead to inequality in society, with the associated range of adverse health and social consequences.

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Section Four: Ethics Section

Word Count – 5,633
(excluding title page and appendix)

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Prepared for: Journal of Health Psychology

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: Navigating the social world with neck dystonia: An interpretative phenomenological analysis.

Name of applicant/researcher: Melanie Maxwell-Scott

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

2. Contact information for applicant:

E-mail: m.maxwell-scott@lancaster.ac.uk **Telephone**: 07918 911 668 (please give a number on which you can be contacted at short notice)

Address: Doctorate in Clinical Psychology, Furness Building, Lancaster University, Lancaster LA1 4YG

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

Dr Fiona Eccles (Lecturer, Doctorate in Clinical Psychology, Lancaster University)

Dr Fiadhnait O'Keeffe (Principal Clinical Neuropsychologist, St Vincent's University Hospital, Dublin).

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 Fiona Eccles (Lecturer, Doctorate in Clinical Psychology, Lancaster University)

Dr Fiadhnait O'Keeffe (Principal Clinical Neuropsychologist, St Vincent's University Hospital, Dublin).

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

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

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?

Data will be obtained through semi-structured interviews.

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

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

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? n o

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? n o

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):

Neck dystonia is a condition that causes stiff, jerking movements of the head and neck. The condition can have a big impact on people's lives, making everyday tasks difficult. Evidence shows people who have visible differences such as this can experience negative views and unfair treatment from others and can feel embarrassed and self-conscious when they are around other people.

This thesis will look at how dystonia effects people's relationships with friends, family and work colleagues. I will interview between eight and 12 people to gain a deep understanding of whether they have experienced negative views from others and felt embarrassed and self-

conscious. This understanding could help clinical psychologists when working with people with neck dystonia, and their families, helping them to cope and adjust to the condition. This thesis also aims to improve public understanding of how visible conditions affect people, raising awareness to reduce unfair treatment in future.

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

Start date: October 2021 End date: March 2023

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

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

8 to 12 participants will be recruited. They will be people of any gender, English-speaking, between the ages of 35 and 65. They must self-report having a diagnosis of idiopathic neck dystonia for at least a year.

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 (eg adverts, flyers, posters).

A recruitment poster will be shared by Dystonia UK (https://www.dystonia.org.uk/) – the UK's largest charity for people with dystonia. They can share this wherever they feel is appropriate and this could include their Facebook page, Twitter feed, mailing lists and newsletters. The poster will invite interested individuals to make contact directly with the student researcher via email.

If this recruitment avenue does not generate a sufficient number of participants, the researcher will set up a project specific Twitter account to circulate the same advertisement. If this second option does not generate sufficient participant numbers, the researcher will include recruitment via Dystonia Ireland, which would proceed similarly to that for Dystonia UK.

Potential participants will then be emailed the Participant Information Sheet and Consent Information Sheet. They will be encouraged to read the material and ask any questions before deciding to take part.

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

The study will be qualitative and use an interpretative phenomenological analysis (IPA) methodology. A sufficient evidence base does not exist regarding the nature of social interactions for people with neck dystonia. Therefore, a qualitative approach is appropriate as an explorative first step in order to understand the topic in greater depth. IPA is appropriate as it is in-depth and can capture the interaction of social and psychological factors. IPA was originally designed to understand the experience of people with chronic conditions (Smith et al, 1999).

There are different ways of analysing the data and presenting the findings of an IPA study. The researcher intends to follow the approach as outlined by Murray and Wilde (2020). This involves transcribing the audio data verbatim, then working transcript by transcript coding data relevant to the research question into themes, keeping as close to the participants' words as possible. Once the themes have been developed for each participant, themes will be compared across participants, to develop over-arching themes for the whole dataset. Evidence from at least three or four participants should support each theme, as outlined by Murray and Wilde (2020).

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.

Participants will have the option of having the interview via a phone call or Microsoft Teams. Phone calls will be recorded directly onto the researcher's computer using the QuickTime Player app. This means the recordings can be immediately transferred to Lancaster University's One Drive. Video recorded interviews will be recorded using the function on MS Teams. A file will be available after the interview which can immediately be transferred to One Drive. Recordings will be stored securely on One Drive, or an equivalent, university-approved, secure cloud service, until the completion of the project.

Interviews will be transcribed by the researcher into Microsoft Word documents, or possibly by using the transcribe function of Teams, Word or other software.

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

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.

It is not anticipated that USB drives will be required. Data will be encrypted by in-built Microsoft Teams software and stored immediately in Lancaster University's One Drive, or an equivalent, university-approved cloud storage.

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

Participants will have the option of having the interview via a phone call or Microsoft Teams. Phone calls will be recorded directly onto the researcher's computer using the QuickTime Player app. This means the recordings can be immediately transferred to Lancaster University's One Drive. Video recorded interviews will be record function on MS Teams. A file will be available after the interview which can immediately be transferred to One Drive. Recordings will be stored securely on One Drive, or an equivalent, university-approved, secure cloud service, until the completion of the project. Audio/video files of the main interview will

be destroyed after the project has been examined but audio consent data will be stored securely for 10 years with the DClinPsy research coordinator

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?

Transcripts will be stored for 10 years by the DClinPsy research coordinator. The data will not be made publicly available under PURE or equivalent. This is because neck dystonia is not a common condition, so even if the data is anonymous it may be possible to identify participants from the interview transcripts.

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

Data will only be made available to authentic researchers on reasonable request.

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?

When responding to the research invitation, the researcher will email potential participants the Participant Information Sheet and Consent Information Sheet. They will be encouraged to read the material and ask any questions before deciding to take part.

The consent form will be read aloud to the participant at the start of the interview, and participants will be audio-recorded giving verbal consent. This recording will be kept separate from the main interview.

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

Discussing issues related to relationships and social interactions may cause some distress for participants. It will be made clear to them at the start that they can stop the interview at any time for a break or for the interview to be rescheduled and the researcher will remain alert to possible signs of distress. Contact details for support services including the Samaritans will be provided on the debrief sheet.

Participants can withdraw their involvement before or during the interview. Participants have up to two weeks after the interview to withdraw their data should they want to. They will be informed of this when they give their consent to participating in the research.

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 safety concerns as the aim is to conduct all interviews from the researcher's home via telephone or video calls. If the researcher experiences distress in relation to anything discussed in the interview, they will in the first instance, discuss this with the project supervisors.

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 may be no direct benefits to participants as a result of this research. Participants may find contributing to the study to be a positive experience because they may find it interesting to reflect on how their lives have changed because of their condition. They may also appreciate contributing to research that aims to improve psychological treatment for people with neck dystonia.

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

None anticipated.

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.

All participant information and interview recordings will be stored confidentially. If during the interview the researcher is concerned about harm coming to the interviewee or others, then the researcher will discuss this information with the project supervisors and may then have to take appropriate action.

All identifiable information will be removed from the transcripts. No identifiable data will be stored with transcripts or the recordings. Any quotations used in the reporting stage will be free of identifiable data as far as possible.

Participants will be asked to pick a pseudonym for the final report. The researcher will recommend they pick a name that reflects their age cohort (as this will help readers of the thesis to imagine the participants' life stage).

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

An expert by experience has been consulted on the overall design on the project and the proposed interview schedule. Changes were made following feedback.

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

Should it be requested, the researcher will send electronic copies of the final, post-viva, passed version of the thesis to participants and Dystonia UK (and if appropriate, Dystonia Ireland). They will present the findings at the University thesis presentation day and publish the thesis online via the University. The aim is also to publish in a specialist health psychology journal, for example, Disability and Rehabilitation. It may also be presented at conferences and special interest groups.

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?

None

SECTION FOUR: signature

Applicant electronic signature:

Date 11/08/2021

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 5/8/2021

Submission Guidance

1. **Submit your FHMREC application by email to Becky Case (**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. **<u>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</u>**

Research protocol

Title: Navigating the social world with neck dystonia: An interpretative phenomenological analysis

Applicant: Melanie Maxwell-Scott

Supervisors: Dr Fiona Eccles (Lecturer, Doctorate in Clinical Psychology, Lancaster University);

Dr Fiadhnait O'Keeffe (Principal Specialist Neuropsychologist, St Vincent's University Hospital, Dublin).

Introduction

What is neck dystonia?

Neck dystonia, also known as cervical dystonia, is the most common form of adult-onset, focal dystonia, with a worldwide prevalence rate estimated at 0.8 per 100,000 (Albanese et al. 2013; LeDoux et al. 2016). However, this could be an underestimate, as dystonia is difficult to diagnose (Albanese, Di Giovanni, & Lalli, 2019). This chronic movement condition is characterised by sustained or intermittent muscle contractions leading to unusual postures of the head and neck, tremors, stiffness and pain (Albanese et al., 2013). Although there are medical treatments such as surgery and botulinum toxin injections which can alleviate

symptoms, there is currently no cure and daily life can be considerably impaired by the condition (Contarino et al., 2016)

Why are social interactions important to study in relation to neck dystonia?

There is limited evidence available as to how people with neck dystonia experience the social world. It is anticipated that difficulties with social interactions will be experienced, particularly because people with neck dystonia experience stigma due to having a visible condition (Papathanasiou, et al., 2001).

Goffman (1963) described a stigmatised individual or group as possessing features which differ from a social norm, either due to physical differences, character flaws or identification with a particular group on the grounds of, for example, race or religion. One study found 51% of participants with neck dystonia reported the feeling of stigmatisation. This was particularly prevalent for those younger than 60 years of age (60% of those under 60 vs. 38.5% of those over 60) (Klingelhoefer et al., 2020).

However, there is limited and mixed evidence on how stigma affects the wellbeing of people with neck dystonia. One study found a significant relationship between stigma and both the mental and physical aspects of quality of life (QoL) in a survey of 289 people with neck dystonia (Ben-Shlomo, Camfield, & Warner, 2002). However, another study of people with segmental dystonia, affecting two or more parts of the body in areas adjacent or close to each other, found no significant relationship between stigma and health related QoL (Basurović et al., 2012). Both studies were limited because stigma was measured using a questionnaire

designed for people with rectal cancer (Macdonald & Anderson, 1984), which has not been validated for use with people with dystonia.

Qualitative research has been used to study the impact of stigma on people with other chronic movement disorders. For example, in one study participants with Parkinson's disease described how they felt embarrassed by their inability to perform physical tasks, and how they percieved others to be feeling sadness, pity and resentfulness towards them due to their condition (Chiong-Rivero et al., 2011).

The experience of stigma is closely related to a person's development and maintenance of social identities. For example, two themes emerged from another study focused on an individual with Parkinson's disease; day-to-day difficulties and a distorted sense of self (Bramley & Eatough, 2005). Social situations exacerbated a feeling that the body is misrepresenting the self. The participant's preferred identity was as someone who is "young at heart" but when people saw her physical difficulties she worried they saw an old and incapable person. Ways to mitigate the impact of stigma on wellbeing for people with Parkinson's disease have been found to include accepting a new social identity and actively managing disclosure of the condition to other people (Hermanns, 2013).

Studying social interactions, both familiar and non-familiar, allows the researcher to explore whether participants have experienced stigma, how it has impacted them, and if they have adapted their social identities in response. This may have a particular impact on middle-aged people (35 – 65 years old), who are likely to have well developed identities in a professional, parenting and/or relational realm and may find it more difficult to modify their identities (Lachman, 2004; Staudinger & Bluck, 2001). Taking a qualitative approach allows the

researcher to understand the issue in more depth, including nuances and complexities of social interactions and relationships.

The importance to clinical psychology

Little is known about how people with neck dystonia experience the social world, and how they may be affected by stigma and changes to social identity. More research in this area will help clinical psychologists and other healthcare professionals to tailor care provision for this group. This may mean using the therapeutic process to help an individual change and adapt their perceptions about themselves and their condition. Deeper understanding could support a multidisciplinary approach to treatment by emphasising the psychological impacts of living with neck dystonia. Systemic interventions may also be required to reduce stigmatising attitudes, such as educational campaigns.

Consequently, this study aims to address the question: What are the social experiences of people with neck dystonia?

Method

Design

A sufficient evidence base does not exist regarding the nature of social interactions for people with neck dystonia. Therefore, a qualitative approach is appropriate as an explorative first step in order to understand the topic in greater depth. The study will follow an interpretative

phenomenological analysis (IPA) approach (for example, as outlined by Smith, et al., 1999). IPA is suitable as it is in-depth and can capture the interaction of social and psychological factors. The method was originally designed to understand the experience of people with chronic conditions (Smith, et al., 1999). Semi-structured interviews will be conducted with each participant. Interviews will be recorded and transcribed by the researcher.

An expert by experience has been consulted on the overall design on the project and the proposed interview schedule. Changes were made following feedback.

Participants

Participants will be adults who self-report having a diagnosis of idiopathic neck dystonia for at least a year. They will be people of any gender, English-speaking, between the ages of 35 and 65. People can take part regardless of relationship status, parental and work status, severity of symptoms, and whether undergoing treatment.

As the aim of an IPA study is to use a small, well-defined, purposive sample, between eight and 12 participants will be recruited (Smith, Flowers, & Larkin, 2009).

Procedure

A recruitment poster will be shared by Dystonia UK (https://www.dystonia.org.uk/) – the UK's largest charity for people with dystonia. They can share this wherever they feel is appropriate and this could include their Facebook page, Twitter feed, mailing lists and newsletters. The poster will invite interested individuals to make contact directly with the student researcher via

email. The advertisement on social media will make it clear that the direct messaging function is to be avoided as it is not secure.

If this recruitment avenue does not generate a sufficient number of participants, the researcher will set up a project specific Twitter account to circulate the same advertisement. The direct messaging function will be turned off to avoid people sharing personal information. If this second option does not generate sufficient participant numbers, the researcher will include recruitment via Dystonia Ireland, which would proceed similarly to that for Dystonia UK.

Potential participants who get in touch will then be emailed the Participant Information Sheet and Consent Information Sheet (see appendices 1,2 and 3). They will be encouraged to read the material and ask any questions before deciding to take part.

Once participants declare they are happy to take part in the study, interviews will be arranged at a convenient time. Due to physical difficulties, the means of interview can be decided by the participants, either by phone call or video call via MS Teams.

The consent form will be read aloud to the participant at the start of the interview, and participants will be audio-recorded giving verbal consent. The rest of the interview will then be audio or video recorded for transcription purposes.

The topic guide (appendix 6) will include areas designed to explore participants' experiences of how living with neck dystonia has affected their social interactions with familiar and non-familiar people. Interviews will last approximately an hour.

Following the interview, participants will be emailed a debrief sheet which includes details of support services in the case of any distress being prompted by the interview.

Participants will be asked if they would like to receive a final, post viva copy of the research project by email. If so, email addresses will be stored separately from other data to protect participants' confidentiality and will be deleted once they have been sent a copy.

Proposed analysis

The data will be analysed using the IPA approach, for example, as outlined by Murray and Wilde (2020). This involves transcribing the audio data verbatim, then working transcript by transcript coding data relevant to the research question into themes, keeping as close to the participants' words as possible. Once the themes have been developed for each participant, they will be compared across participants, to develop over-arching themes for the whole dataset. Evidence from at least three or four participants should support each theme (Murray & Wilde, 2020).

Practical issues

Confidentiality

All participant information and interview recordings will be stored confidentially (see below for further information). The only time confidentiality may need to be broken is if during the interview the researcher is concerned about harm coming to the interviewee or others. In this case, the researcher will discuss this information with the project supervisors if possible and take appropriate action (for example, contact the police or social services).

Anonymity

All identifiable information will be removed from the transcripts. No identifiable data will be stored with transcripts. Any quotations used in the reporting stage will be free of identifiable data as far as possible.

Participants will be asked to pick a pseudonym for the final report. The researcher will recommend they pick a name that reflects their age cohort (as this will help readers of the thesis to imagine the participants' life stage).

Distress management

Discussing issues related to relationships and social interactions may cause some distress for participants. It will be made clear to them at the start that they can stop the interview at any time for a break or for the interview to be cancelled or rescheduled. Contact details for support services including the Samaritans and Dystonia UK (or Dystonia Ireland where appropriate) will be provided on the participant information and the debrief sheet.

Data storage

Participants will have the option of having the interview via a phone call or Microsoft Teams. Phone calls will be recorded directly onto the researcher's computer using the QuickTime Player app. This means the recordings can be immediately transferred to Lancaster University's One Drive. Video recorded interviews will be recorded using the function on MS

Teams. A file will be available after the interview which can immediately be transferred to One Drive. Recordings will be stored securely on One Drive, or an equivalent, university-approved, secure cloud service, until the completion of the project.

Interviews will be transcribed into Microsoft Word documents by the researcher, possibly by using the transcribe function of Teams, Word or other software.

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

Withdrawal of consent

Participants have up to two weeks after the interview to withdraw their data should they want to. They will be informed of this when they give their consent to participating in the research.

Safety of researchers

There are no anticipated safety concerns as the aim is to conduct all interviews from the researcher's home via telephone or video calls.

Timescale

Ethics application: August 2021

Proposed Ethics approval: October 2021

Recruitment: November 2021 – June 2022

Analysis and write up: June 2022 – February 2023

Submission: March 2023

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Ethics Committee Approval Letter

Applicant: Melanie Maxwell-Scott
Supervisor: Dr Fiona Eccles, Dr Fiadhnait O'Keefe
Department: DHR
FHMREC Reference: FHMREC20195

13 October 2021

Re: FHMREC20195
Navigating the social world with neck dystonia: An interpretative phenomenological analysis

Dear Melanie,

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.

Email: fhmresearchsupport@lancaster.ac.uk

Yours sincerely,

T. Morley

Tom Morley,
Research Ethics Officer, Secretary to FHMREC.

Appendix 4 – A: Topic guide

As this is a qualitative study following the approach of interpretative phenomenological analysis, a topic guide will be used to guide the interviews while allowing freedom for the researcher to explore more deeply what the interviewees raise.

Topic 1

Background information on life circumstances and introduction, such as:

- how long they have had neck dystonia
- how their dystonia affects their movement
- brief story of how dystonia manifested, the route to diagnosis
- treatment and care management
- close relationships, including partners, family and friends
- living arrangements and whether they have caring responsibilities
- working life and occupation
- hobbies and interests.

Topic 2

The nature of social relationships and whether and how these have changed since having neck dystonia, including interactions with:

- family
- friends
- work colleagues
- acquaintances
- unfamiliar people.

Topic 3

The context to social relationships and whether this impacts on interactions, for example:

- being at home
- other people's homes
- restaurants, cafes and pubs
- places of religion, support groups
- travelling on public transport
- hospital and clinics
- the virtual world and social media.

Topic 4

Reflections on what these experiences mean to the person as an individual, for example:

- personality changes, feeling more or less sociable

- lessons they have learnt about themselves
- broadening or shrinking of social circle
- attitudes towards other people
- ways of coping with the condition or adapting to a new way of living
- coping with the diagnosis given that dystonia has no cure
- advice they would give to people newly diagnosed with neck dystonia.

Appendix 4 – B: Participant information

For further information about how Lancaster University processes personal data for research purposes and your data rights please visit our webpage: www.lancaster.ac.uk/research/data-protection

Participant information

Navigating the social world with neck dystonia: An interpretive phenomenological analysis

Thank you for your interest in this research project. My name is Melanie Maxwell-Scott and I am conducting this research as a student on the Clinical Psychology Doctorate programme at Lancaster University, Lancaster, United Kingdom.

What is the study about?

The purpose of the study is to understand how social interactions are experienced from the perspective of a person with neck dystonia. I am interested in how your experience of neck dystonia has impacted on interactions with family, friends, work colleagues, professionals and/or general people you meet. Has it made social connections harder or easier to develop and maintain? Have you changed the way you act around other people since having neck dystonia? Have you experienced any difficulties with others/ with the public due to your neck dystonia?

Why have I been approached?

You have been approached because we would like to interview people who are **between 35 and 65 years old** and who have a diagnosis of neck dystonia/ cervical dystonia / spasmodic torticollis for at least a year.

Do I have to take part?

No. It is up to you whether you decide to take part in this research. You can withdraw your involvement at any time; before, during, or up to two weeks after the interview.

What will I be asked to do I take part?

If you decide to take part in the research, you will be contacted by the researcher to arrange an interview at a time convenient for you. The interview will take up to an hour. It will be conducted by phone or video call. The interviewer will ask about if and how your social relationships have changed since having a diagnosis of neck dystonia and what your feelings are about this.

Will my data be identifiable?

The recordings of interviews collected for this study will be stored securely on a University approved secure cloud storage, and only the researchers conducting this study will have access to these recordings:

- Audio/ video recordings of the main interview will be deleted once the project has been submitted for publication/examined.
- The files on the computer will be encrypted (that is no-one other than the researcher will be able to access them) and the computer itself password protected.
- The typed version of your interview will be made anonymous by removing any identifying information including your name. Anonymised direct quotations from your interview may be used in the reports or publications from the study, so your name will not be attached to them. All reasonable steps will be taken to protect the anonymity of the participants involved in this project.
- All your personal data will be confidential and will be kept separately from your interview responses.
- Transcripts and audio recordings of the consent process will be kept securely for ten years and then deleted.

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

What will happen to the results?

The results will be summarised and reported in a thesis and may be submitted for publication in an academic journal. A report of the findings will also be shared with you and the other interviewees (should you want to see it) and with Dystonia UK.

Are there any risks?

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

Are there any benefits to taking part?

Although you may find participating interesting, there are no direct benefits in taking part. There may be indirect benefits to taking part, as the research aims to increase understanding of dystonia and to contribute to better treatment and management of the condition.

Who has reviewed the project?

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

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

If you have any questions about the study, please contact the main researcher:

Melanie Maxwell-Scott

Email: m.maxwell-scott@lancaster.ac.uk

You can also contact the research supervisor:

Dr Fiona Eccles

Email: f.eccles@lancaster.ac.uk

Tel: 01524 592 807

Complaints

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

Dr Ian Smith, Tel: +44 (0)7507 857069

Research Director for Clinical Psychology Email: I.smith@lancaster.ac.uk

Division of Health Research, Faculty of Health and Medicine

Lancaster University

B31 Floor, Health Innovation One

Sir John Fisher Drive

Lancaster University

Lancaster

LA1 4AT

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

Dr Laura Machin Tel: +44 (0)1524 594973

Chair of Faculty of Health and Medicine Research Ethics Committee

Email: l.machin@lancaster.ac.uk

Faculty of Health and Medicine

(Lancaster Medical School)

Lancaster University

Lancaster

LA1 4YG

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

Resources in the event of distress

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

Your GP: we recommend speaking to your own hospital consultant or GP for support.

Samaritans

www.samaritans.org

Tel: 116 123

Email: jo@samaritans.org

Dystonia UK

www.dysonia.org.uk

Tel: 020 7793 3651

Email: support@dystonia.org.uk

Appendix 4 – C: Research invitation

- Have you had a diagnosis of neck dystonia (cervical dystonia or spasmodic torticollis) for at least a year?
- Are you between the ages of 35 and 65?
- Could you spare one hour to be interviewed about your experiences of living with the condition?

My name is Melanie Maxwell-Scott, and I am a student on the Clinical Psychology doctoral course at Lancaster University. This project aims to better understand how living with neck dystonia impacts on social interactions with family, friends and work colleagues and on an individual's social identity. I am interested in your thoughts and feelings as to how relationships with others may have changed since your diagnosis of neck dystonia.

If you are interested in participating and would like further information, please get in touch:

☎ Phone: +44 (course mobile phone number to be inserted)

✉ Email: m.maxwell-scott@lancaster.ac.uk

Please do not use the direct messaging function as this may not be secure.

Project supervisor: Dr Fiona Eccles, Lecturer, Doctorate in Clinical Psychology, Lancaster University.

Researcher: Melanie Maxwell-Scott, Trainee Clinical Psychologist, Lancaster University.

Appendix 4 – D: Consent information

Study Title: Navigating the social world with neck dystonia: An interpretative phenomenological analysis.

Please note you do not need to fill in this form or post it back. It is just for information. The researcher will go through each point during the interview and you will be able to give verbal consent.

We are asking if you would like to take part in a research project looking at how social interactions are impacted by neck dystonia and whether your relationships with family, friends and/or colleagues have changed.

If you have any questions or queries regarding this information before the interview before please speak to the principal investigator, Melanie Maxwell-Scott.

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

2. I confirm that I have had the opportunity to ask any questions and to have them answered.

3. I understand that my interview will be audio (or video) recorded and then made into an anonymised written transcript.

4. I understand that audio (or video) recordings will be kept until the research project has been examined.

5. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without my medical care or legal rights being affected.

6. I understand that after the interview I have up to two weeks to change my mind and withdraw consent for my data to be used.

7. I understand that the information from my interview will be combined with other participants' responses, anonymised and may be published; all reasonable steps will be taken to protect the anonymity of the participants involved in this project.

8. I consent to information and quotations from my interview being used in reports, conferences and training events. These will also be anonymous.

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

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

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

Name and date

Appendix 4 – E: Participant debrief information

Thank you for participating in this study and sparing your time to contribute to research.

The purpose of the study is to gain a deeper understanding of the social experiences of people with neck dystonia. In particular, we want to understand whether relationships with family, friends, colleagues and acquaintances have changed and in what way. We are anticipating there could be difficulties with social interactions for some people as neck dystonia physically affects how people communicate. The condition is visible to others which may mean stigma is experienced. As dystonia tends to occur in mid-life, people may find they have to develop a new identity, and this can sometimes be difficult to manage.

This information will help clinical psychologists and other professionals understand some of the social pressures that people with neck dystonia experience. This may help to tailor the care provided. The research also aims to raise general public awareness of neck dystonia in order to reduce any potential stigma or misunderstanding about the condition.

The interview will be anonymously transcribed into a word document. It will be consolidated with other interview transcripts so the researcher can look for common themes across the experience of interviewees. These themes will be written up in a report and published by Lancaster University. The researcher hopes to submit the final report for inclusion in an academic journal.

If you would like, the researcher will email or post a copy of the final report to your address. If you have any questions about the research or the process, please contact m.maxwell-scott@lancaster.ac.uk

If you have experienced any distress from things discussed during the interview, or at any point in the future, then the following resources may be helpful:

Your GP: we recommend speaking to your own hospital consultant or GP for support.

Samaritans

www.samaritans.org

Tel: 116 123

Email: jo@samaritans.org

Dystonia UK

www.dysonia.org.uk

Tel: 020 7793 3651

Email: support@dystonia.org.uk