Title: Lived Experiences of Psychological Distress in Neck Dystonia

Short title: *Psychological Distress in Neck Dystonia*

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Abstract:

Purpose: Neck dystonia (ND) is a hyperkinetic movement disorder affecting the head and neck regions. High prevalence rates of psychological distress have been reported in ND. The aim of this research was to explore people's experiences of distress, in order to understand the needs of this population further.

Materials and Methods: Interpretative phenomenological analysis (IPA) was used to analyse data obtained from 11 semi-structured interviews with people with ND, recruited through a UK based charity.

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

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

Keywords:

Cervical dystonia, spasmodic torticollis, wellbeing, mood, distress

Data availability statement:

The data that support the findings of this study are available from the corresponding author upon reasonable request.

No funding sources to acknowledge. The authors report there are no competing interests to declare.

Statement of Contribution

What is already known on this subject?

Existing literature demonstrates that there is a high prevalence rate of psychological distress in people with neck dystonia. Some researchers believe that the distress arises from living with the debilitating symptoms of dystonia, whilst other researchers suggest that distress is intrinsic to the neurobiological processes of the condition.

What does this study add?

This study offers a novel perspective to the ongoing debate regarding psychological distress in neck dystonia, by examining how individuals themselves experience and understand their distress. Most people who took part in this study believed that psychological difficulties came from living with challenging symptoms, with some wondering whether distress preceded the onset of the condition.

Introduction

Dystonia is a hyperkinetic movement disorder characterised by sustained or intermittent muscle contractions, resulting in abnormal and often painful movements and postures (Di Biase et al., 2022). Adult-onset idiopathic focal dystonia (affecting a specific body region) is the most common form, of which neck dystonia (ND), or cervical dystonia, is the most common phenotype (Ben-Shlomo et al., 2002). The condition is associated with uncontrollable spasmodic movements, which can cause the neck to rotate, flex, extend, tilt or shift laterally (Velickovic et al., 2001). The involuntary movements that characterise this condition are associated with loss of inhibition within certain brain circuits including the brainstem, basal ganglia, cerebellum and cortex (Albanese et al., 2023). While there is no cure for the condition, symptoms may be alleviated using botulinum toxin intramuscular injections, typically delivered every three months (Moll et al., 2018). Deep brain stimulation

(DBS) may be considered as an alternative treatment in severe cases of dystonia and involves the surgical insertion of electrical stimulators to control brain signals and reduce involuntary movements. (Rodrigues et al., 2019).

While ND has been considered predominantly a motor disorder for many years, there has been growing interest in recent decades in the non-motor aspects of the condition, in particular the psychological component (Costanzo et al., 2021). Results from prevalence studies indicate that anxiety and depression are present in approximately 25 to 60 % of people with ND (Ceylan et al. 2019; Comella & Bhatia, 2015; Müller et al., 2002; Ndukwe et al., 2020; Tomic et al., 2016). A study examining 201 individuals with ND found that pain, disability and mood were significant factors in reducing a person's quality of life (QoL), with mood being reported as the most predominant predictor (Ndukwe et al., 2020). Furthermore, stigma arising from postural abnormalities and changes in body image (Papathanasiou et al., 2001; Paracka et al., 2020) has been found to have a negative impact on personal, social and professional domains of QoL (Tomic et al., 2016).

Some researchers believe that mood difficulties are a primary symptom of the condition, intrinsic to the neurobiology driving the motor difficulties, while others believe that mood difficulties are secondary to living with the debilitating symptoms. In support of the former argument, studies have found that approximately 43% of people with ND experience mood difficulties prior to onset of motor symptoms (Moraru et al., 2002; Wenzel et al., 1998), suggesting that distress is a primary symptom of the condition. Further evidence comes from a study by Lencer et al. (2009), examining 86 individuals with focal dystonia, the majority of whom had ND. The findings reported a mean age of 24.3 years for mood difficulties and 42.5 years for motor symptoms. Recent longitudinal research has demonstrated stability of mood difficulties despite changing severity of motor symptoms, indicating that distress is an independent component of the condition (Berardelli et al., 2015).

On the other side of this debate, Mueller et al. (2008) reported that mood improved with successful treatment of the physical symptoms, indicating that psychological distress is likely to be secondary to living with ND. Similarly, Skogseid et al. (2007) reported an association between severity of symptoms and low mood. Psychosocial variables such as body image, stigma, illness perceptions, self-esteem and cognitive ability have all been reported to be associated with psychological distress (Gowling et al., 2024; Jahanshahi, 1990; Monaghan et al., 2022; O'Connor et al., 2023a) and QoL (Ben-Shlomo et al., 2002; Tomic et al., 2016). Currently, little is known about the perspectives of individuals with ND regarding this debate.

Currently, treatment plans tend to prioritise the medical needs of this group, with other aspects of the condition typically being neglected (Ndukwe et al., 2020). A systematic review into the effectiveness of behavioural therapies in the management of dystonia was unable to draw any clear conclusions due to limited research in this area (Bernstein et al., 2016). The National Institute for Health and Care Excellence (NICE) guidelines (2009) currently recommend a Cognitive Behavioural Therapy (CBT) approach to managing psychological distress in people with long-term health conditions. However, this pathway is based on recommendations developed for non-dystonic populations, and therefore does not take into account the specific needs of this particular group (Zurowski et al., 2013). The majority of research investigating psychological distress in ND have been quantitative in design, with few studies exploring the lived experiences of this population. A recent study by Morgan et al. (2019) used phenomenological methods to explore the experiences of people with different types of dystonia. Their results indicated that the psychological aspect of ND was particularly salient for participants, with recommendations for future phenomenological studies to focus on one particular type of dystonia.

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

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

Materials and Method

Design

Interpretative phenomenological analysis (IPA) was the methodological approach chosen for this study. Unlike more descriptive qualitative approaches, the phenomenological element of IPA focuses not just on a person's experience of what happened, but on the meaning they place on that experience. This, along with the double hermeneutic process of interpretation, and the focus on the particular, allows IPA research to understand the perspective of the person, as much as possible (Smith et al., 2022). In line with published guidelines regarding service user involvement in clinical psychology research, an individual with lived experience of ND was consulted throughout the design process (Sheldon & Harding, 2010). They provided input regarding terminology, interview schedule and supporting documentation. Ethical approval for this research was granted by the faculty ethics committee at the research's institution (FHM-2023-1010-SA-1).

Participants

Participants were selected through the Dystonia UK charity. Participants were selected using purposive sampling, a technique widely used within qualitative research to identify individuals that are knowledgeable and experienced in the specific research question

(Palinkas et al., 2015). Individuals aged 18 to 69, with adult-onset ND, were included. Individuals with other significant concurrent health difficulties affecting daily functioning were excluded, to ensure homogeneity of the sample (Smith et al., 2022). Individuals must have been living with ND for at least six months to allow for a period of adjustment. Inclusion criteria specified that participants experienced psychological distress either currently, since their diagnosis, or up to six months prior to diagnosis. The study as advertised on the charity website and newsletter, and eligible participants were invited to contact the researcher directly to express their interest in participation. The final sample of 11 participants consisted of six females and five males, ranging in age from 58 to 69. Table 1 outlines the characteristics of the sample. Pseudonyms have been used to ensure anonymity and were chosen by the researcher to reflect the age, gender and culture of participants.

Table 1

Participant Demographics

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

Data Collection

Semi-structured interviews were used to collect the data. Following recommendations by an expert by experience, simple language was used to maximise accessibility and inclusivity. The interview schedule (Table 2) opened with a question that asked participants to recall the lead up to their diagnosis. The purpose of this question was to encourage a descriptive response that would allow the participants to settle into the interview setting. As the interview progressed, the interviewer guided participants to discuss the impact of ND on daily living, their experiences and understandings of psychological distress, and support for distress. Topics appearing salient to participants were explored more fully. Call recording software was used during the interviews. Seven interviews took place via video, and four by telephone, with participants choosing their preferred method to ensure comfortableness during the process. The interviews ranged in duration from 40 to 68 minutes and were transcribed verbatim.

Table 2

Semi-Structured Interview Schedule

Topic 1-Experiences of Neck Dystonia
Can you describe your journey to getting a diagnosis of neck dystonia?
-How long have you been living with neck dystonia?
-Can you describe for me the symptoms of neck dystonia?
-What symptoms are most prominent for you?
Topic 2-Impact on Quality of Life
-How does neck dystonia impact your daily life (e.g., family, friends, work/not being able to work)
-What has been most difficult for you about this condition/what are the main challenges?
-Has neck dystonia stopped you from doing anything that you would like to do?
Topic 3: Experiences and Understanding of Psychological Distress
-When we talk about psychological distress we are referring to feelings like anxiety, depression and stress.
-Can you tell me about any experiences of psychological distress?
Did you have any psychological difficulties prior to diagnosis?

-What do you think has contributed to distress for you?

-Some researchers say that mood difficulties are as much a symptom of neck dystonia as motor symptoms. Other researchers believe that mood difficulties happen as a result of living with the movement symptoms. How do you understand psychological distress in neck dystonia?

Topic 4: Support for Psychological Distress

-Have you ever received psychological support for anxiety, depression or stress?

-Have you talked to your neurologist about how your mood is affected? How have these conversations gone? What support or treatment was recommended for you?

-Have you ever been prescribed medication for your mood? How do you find the medication?

-What do you find is most helpful to you in managing any psychological difficulties that you have?

-How have family/friends/work responded to psychological distress you have experienced?

-Do you think you receive sufficient psychological support or do you think there is a need for more?

Anything else?

- Is there anything I might have missed that you think might be relevant or helpful for me to know in the context of this research?

Data Analysis

Analysis followed recommendations for conducting IPA by Smith et al. (2022). This involved an iterative process of reading, re-reading and exploratory noting, and the construction of descriptive and interpretative experiential statements. In line with an idiographic approach, each transcript was analysed fully before the researcher moved onto the next one. Once each participant had a personal experiential table, the final stage involved the development of group experiential themes (GET's), identifying shared and unique experiences of psychological distress. To ensure that this research was of high quality and validity, the researcher referred to the core principles of qualitative research, outlined by Yardley (2000): sensitivity to context, commitment and rigour, transparency and coherence, impact and importance. The researcher continually evaluated their own assumptions or biases throughout the interpretative process through use of a reflective journal, detailing each stage of the analytical process (Vicary et al., 2017). To ensure transparency of the process, the researcher included a statement about the characteristics of the research team (Levitt et al., 2018).

Researcher Statement

This research was conducted as part of a clinical psychology doctorate; therefore the main researcher was skilled in managing distress, and creating a comfortable space for participants discuss emotive topics. Considering sensitivity of context (Yardley, 2000), they consulted with an expert by experience during the study design stage to ensure that this project was person-centred. The research team also included an experienced clinical psychologist and clinical neuropsychologist to ensure that this research was clinically and academically relevant.

Results

The analysis produced four GETS, representing the overarching themes within the data: 1) Uncertainty in the lead up to diagnosis; “I was so full of fear” 2) The isolating nature of the condition; “you don’t know who to turn to” 3) Mind and body relationship; “is my head turning because I’m thinking about it?”, and 4) A challenging new way of life; “living life sideways”.

Uncertainty in the lead up to Diagnosis; “I was so full of fear”

There was a sense that the journey to diagnosis was long, and shrouded in uncertainty. Participants worried about why they were experiencing their new symptoms, and this anxiety was perpetuated by healthcare services that were at times dismissive. This theme also captures the experiences of ambiguity upon receiving a diagnosis in relation to understanding about the condition.

Worrying about symptoms

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

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

Feeling dismissed by healthcare providers

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

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

Sharon believed that her anxiety came from not knowing why she was experiencing the physical symptoms. It appeared that her understanding of distress was misaligned with that of her doctor’s, who told her that her physical symptoms were a consequence of her anxiety.

Brian had a similar experience of physical symptoms being dismissed as anxiety, which led to feelings of hopelessness, increased psychological difficulty, and the development of unhelpful coping strategies: “I didn't know what was wrong with me. They weren't gonna do anything about it and em, not much I could do about it, so I just resorted to drink and drugs.”

Unrealistic expectations and false hope

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

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

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

Being left to manage the condition alone

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

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

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

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

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

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

Nobody else understands this

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

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

The embarrassment of navigating the social world

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

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

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

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

Curiosity about the role of earlier trauma

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

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

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

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

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

Regaining control of body

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

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

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

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

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

The incessant discomfort brings you down

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

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

Struggling to accept that this is not going away

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

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

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

Grieving for loss of self

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

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

Discussion

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

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

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

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

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

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

Clinical Implications

The study identified that psychological distress remains a neglected yet salient part of living with ND. All participants reported experiences of distress, however none of these individuals had been offered support for such difficulties. Psychologists could support people with ND through a combination of indirect and direct input. Through formulation, psychologists can develop a shared understanding of a person's difficulties with other professionals, with the aim of reducing the feelings of uncertainty and abandonment identified in this study, and supporting more person-centred care (Geach et al., 2018).

The adjustment literature discussed above identified cognitive and behavioural factors associated with poorer adjustment to chronic illnesses, thus suggesting the potential benefits of a CBT approach. Social withdrawal arising from feelings of embarrassment may be perpetuating feelings of low mood or loneliness for individuals, therefore supporting individuals to develop alternative behaviours may help to improve psychological outcome. The present study indicated the salience of shame and self-criticism in ND, identifying such feelings in relation to emotions, body image, and appearance. Compassion focused therapy (CFT) is a therapeutic model that was specifically developed to address feelings of shame and self-criticism, and as such may be well placed to support people to develop a more compassionate relationship with themselves (Gilbert, 2014).

Limitations

ND is a heterogeneous condition, which posed challenging for an IPA study requiring homogeneity of the sample. Differences in severity of symptoms, support networks, and attitudes of healthcare teams may have impacted people's experiences of distress differently. The choice to use purposive sampling to recruit eligible participants through a charity allowed participants to speak openly about their experiences of healthcare systems. However, there were limitation to this, in that we did not have access to patient records and information regarding severity of symptoms. We were also relying on participants' own reports of their diagnosis. The choice not to collect data regarding disease severity may have limited researchers' ability to consider the effect that symptom severity may have had upon distress, and future research may consider a mixed methods approach to consider patterns within the findings.

Disease duration in the current sample ranged from six months to 40 years, thus individuals living with the condition longer may have adjusted to the condition differently to those with a more recent diagnosis. To ensure homogeneity, strict eligibility criteria were employed, meaning that certain voices were not heard in this particular study, including those above working age, and those with concurrent health conditions.

Conclusion

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

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