DOCTOR OF CLINICAL PSYCHOLOGY (DCLINPSY)

Doctorate in Clinical Psychology : Main Research Portfolio

1) How common is fatigue across chronic illness conditions in children 4693 words and young people? A systematic review ;2) Evaluation of a mindful life group: A service-related project ;3) A mixed-methods study of fatigue in young people with neuromuscular conditions: “It limits how much I can do, it limits the quality of how much I can do, and it's a reminder of your disability which [affects] mental health.”

Williams, Kiesha

Award date:
2022

Awarding institution:
University of Bath

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Research Portfolio Submitted in Part Fulfilment of the requirements for the Degree of Doctorate in Clinical Psychology

Kiesha Williams

Doctorate in Clinical Psychology

University of Bath
Department of Psychology

May 2022

Impact of COVID-19 Pandemic on Research

In March 2020, the start of the COVID-19 pandemic, I was only five months into my doctorate training; all three research projects included within this portfolio have therefore been conducted during the pandemic, however only two were affected. The following commentary provides a brief overview of the impact of COVID-19 on two of the three doctoral research projects.

I had planned for my service-related project to be completed within my second placement (a later life team). Unfortunately, this placement changed at the last minute due to staff uncertainty at the start of the pandemic. Fortunately, the data had been routinely collected by the service over several years and, as I was placed within the same NHS trust, I was able to continue with the initial plan. This meant that, other than some brief uncertainty, my completion of the analysis and write up was not impacted significantly. There was an impact on the dissemination process, however, as whilst I was able to meet with the team virtually to share the findings of the project, I wonder if the impact would have been greater if I had been embedded within the team on placement and in a position to help implement recommendations of the project.

My main research project was designed during the pandemic and so did not require changes or adaptations due to COVID-19. The project was designed to involve an online survey with recruitment via charities and so, following ethical approval, this project could start and continue as planned. Recruitment for this project spanned from February 2021 to February 2022, entirely peri-pandemic, and so it is unclear what impact the pandemic had more broadly on recruitment. During qualitative interviews, participants described fatigue and pandemic experiences and so it is likely that this had some impact on findings for this project, although it is difficult to quantify this.

As healthcare workers within the NHS, my external supervisor and I experienced added clinical pressures during the pandemic. These pressures, as well as the pandemic more generally, had an impact on my personal wellbeing as there was less opportunity for social support from friends, family, and peers. Additionally, I experienced COVID-19 during the write up stage of the main research project which also affected my wellbeing and fatigue levels. These factors will undoubtedly have impacted my ability to work on my research portfolio during this time.

Kiesha Williams

University of Bath

Word Counts

Literature Review	
How common is fatigue across chronic illness conditions in children and young people? A systematic review	4693 words
Service-Related Project	
Evaluation of a mindful life group: A service-related project	4996 words
Main Research Project	
A mixed-methods study of fatigue in young people with neuromuscular conditions: “It limits how much I can do, it limits the quality of how much I can do, and it's a reminder of your disability which [affects] mental health”	5906 words
Executive Summary	983 words
Total word count	16578 words

Note. All word counts exclude abstracts, figures, tables, footnotes, and references.

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Literature Review – Abstract

Objective: Fatigue is a common experience for adults with chronic health conditions. Less is understood about the prevalence of fatigue in the paediatric population. This review aimed to synthesise what is known about the point prevalence of fatigue across chronic health conditions within children and young people. **Methods:** Three databases were searched, from January 2000 to July 2021, to identify studies reporting prevalence rates in chronic health conditions in under 18s. Methodological quality was assessed with a modified version of the Newcastle-Ottawa Scale for Cross-Sectional Studies. **Results:** 25 studies were included. Variations in the assessment of fatigue, across a range of conditions, and heterogenous reporting of prevalence precluded meaningful meta-analysis. Discrepancies in prevalence reports were noted within and across conditions and between child and parent reports but showed fatigue to be more prevalent in those with chronic health conditions compared to healthy peers. **Conclusion:** Despite discrepancy in prevalence rates of fatigue, some level of fatigue is present across chronic health conditions and tends to be higher in this population compared to healthy peers. Limitations alongside clinical implications and recommendations for future research are discussed.

Service-Related Project – Abstract

Objectives: This service improvement project evaluated a twelve-week mindfulness group facilitated with people with a range of difficulties across adult and later life secondary mental health teams. The study aimed to evaluate the impact of the group on levels of mindfulness and psychological distress. **Methods:** Participants were those who completed the group (*n*=70) and completed two standardised self-report measures of distress (*n*=38) and mindfulness (*n*=40) which were completed at the beginning and end of the intervention, over a five-year period and with 14 cohorts of the group. **Results:** There was a significant increase in overall levels of mindfulness over the course of the intervention, with a medium effect size, as well as within three specific components of mindfulness - non-reacting, observing and non-judging. A small but non-significant decrease in psychological distress was noted from the start to the end of the group and not reacting to inner experiences is presented as a key mindfulness skill in supporting the reduction of distress. **Conclusions:** Mindfulness-based skills increased over the twelve-week intervention and small reductions in distress were noted. Improvements in psychological distress may be further supported with additional practices which

concentrate on developing a non-reactive stance. The implications for the service, alongside recommendations for future-research, are discussed.

Main Research Project – Abstract

Neuromuscular conditions are progressive disorders associated with muscular weakness. Fatigue is a common symptom in adults with specific types of neuromuscular conditions, however, less is known about fatigue in young people with neuromuscular conditions. This study aimed to explore prevalence and severity of fatigue, and associated factors, alongside lived experiences of fatigue in young people with neuromuscular conditions. A cross-sectional mixed methods design was used; thirty-three participants with ten neuromuscular conditions completed an online survey including measures of fatigue, mood, sleep quality, responses to symptoms, social functioning, physical functioning, and quality of life. Data were analysed with bivariate correlations and stepwise linear regression. Twelve participants completed follow-up interviews to explore fatigue experiences, analysed with Reflexive Thematic Analysis. In total, 24% of participants reported significant fatigue, which was associated with sleep quality ($r = -.39$, $p = .025$) and depression symptoms ($r = .40$, $p = .022$). Five themes were constructed from interviews capturing the far-reaching impact of fatigue and difficulties in managing it. Quantitative measures and qualitative interviews indicate that fatigue is a common experience for young people with neuromuscular conditions and is strongly associated with feelings of depression. Implications for services, alongside limitations of this study and recommendations for future research are discussed.

Literature Review

How common is fatigue across chronic illness conditions in children and young people? A systematic review.

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Proposed Journal: Journal of Psychosomatic Research (Appendix A)
The Journal of Psychosomatic Research publishes research related to psychology and medicine, with particular focus on those with illness or health conditions and welcomes studies concerning children and adolescents. The journal also publishes systematic reviews, with a word limit of 4-5000 words.

How common is fatigue across chronic illness conditions in children and young people? A systematic review.

Abstract

Objective: Fatigue is a common experience for adults with chronic health conditions. Less is understood about the prevalence of fatigue in the paediatric population. This review aimed to synthesise what is known about the point prevalence of fatigue across chronic health conditions within children and young people. **Methods:** Three databases were searched, from January 2000 to July 2021, to identify studies reporting prevalence rates in chronic health conditions in under 18s. Methodological quality was assessed with a modified version of the Newcastle-Ottawa Scale for Cross-Sectional Studies. **Results:** 25 studies were included. Variations in the assessment of fatigue, across a range of conditions, and heterogenous reporting of prevalence precluded meaningful meta-analysis. Discrepancies in prevalence reports were noted within and across conditions and between child and parent reports but showed fatigue to be more prevalent in those with chronic health conditions compared to healthy peers. **Conclusion:** Despite discrepancy in prevalence rates of fatigue, some level of fatigue is present across chronic health conditions and tends to be higher in this population compared to healthy peers. Limitations alongside clinical implications and recommendations for future research are discussed.

Introduction

Fatigue is a subjective experience (Loades & Chalder, 2020), but one that is typically defined as an overwhelming and persistent feeling of tiredness and exhaustion (Dittner et al., 2004), and often associated with a lack of energy which can disrupt daily functioning (de Vries et al., 2010; Ream & Richardson, 1997). In young people, the lifetime prevalence of persistent fatigue is estimated at 1.29-2.34% (Farmer et al., 2004). In adults, the prevalence of fatigue, which is common and disabling (Small & Lamb, 1999), is higher among those with health conditions (Falk et al., 2007; Goërtz et al., 2021) . Chronic health conditions refer to illnesses that are experienced for at least three months, with no cure, and can be lifelong (Law et al., 2019). This includes a broad range of conditions, such as muscular dystrophy, sickle cell disease, and diabetes of which fatigue is reported as a common feature (Ameringer et al., 2014; Bhullar et al., 2018; Segerstedt et al., 2015).

In previous research, fatigue has been described by young people with multiple sclerosis (MS) as a heavy ‘sandbag’-like feeling, like ‘looking through a haze’, which impacts physical, mental and emotional wellbeing, with parents understanding the experience as a debilitating and ‘uncontrollable tiredness’ (Carroll et al., 2016, pp. 940-942). Similar experiences have been reported by young people with chronic fatigue syndrome who describe feeling ‘drained’ and ‘worn out’ (Parslow et al., 2018, p. 2). These descriptions illustrate the multifaceted nature of fatigue, presenting cognitively and physically (Armbrust et al., 2016), and affecting many areas of life, such as learning, school attendance, and relationships with friends and family (Carroll et al., 2016; Crawley & Sterne, 2009; Garralda & Rangel, 2004; Haig-Ferguson et al., 2009; Winger et al., 2014).

Fatigue in those with chronic health conditions may be related to social and environmental triggers (Loades & Chalder, 2020), as well as biological aspects of the condition or treatment (Ameringer et al., 2014). The cognitive behavioural model of fatigue (Surawy et al., 1995) purports that many factors may maintain fatigue, such as cognitive, emotional and behavioural elements, in addition to external reinforcement from others. This model has been supported by further research which shows a relationship between fatigue and a multitude of bio-psycho-social factors, including pain, treatment, low mood, sleep, and performance at school (Armbrust et al., 2016; Carroll et al., 2015; Spathis et al., 2015).

In the paediatric population, previous systematic reviews have considered the prevalence of fatigue within specific health conditions. Armbrust and colleagues (2016), for example, reported fatigue in 60-76% of young people with juvenile idiopathic arthritis. Fatigue was also reported as a concern for young people with MS (Carroll et al., 2015), and in young people with cancer although a lack of relevant studies limited the conclusions that could be drawn on the prevalence and consequences of fatigue for this population (Nowe et al., 2017). Other reviews have explored correlates or contributors to fatigue, such as mood, within young people with chronic conditions (Carroll et al., 2015; Van de Vijver et al., 2019). One such review (Eddy & Cruz, 2007) found a relationship between fatigue and reduced quality of life in young people with chronic health conditions but noted that the majority of participants fell within the paediatric cancer population. This indicates that less is understood about the prevalence rates of fatigue within other health conditions and information from such studies has not yet been synthesised. Furthermore, a recent cross-sectional study (Nap-Van Der Vlist et al., 2019) with young people indicated that fatigue was prevalent across three different chronic conditions, leading authors to recommend the need for a systematic assessment of fatigue within this population. Despite these studies, previous research has not synthesised information on point prevalence of fatigue in children and young people with chronic health conditions and doing so would be valuable to inform support offered by clinical teams.

Point prevalence is the proportion of a population currently experiencing a condition, such as fatigue. This provides an estimate on who within a population experiences a condition, at a certain time point, and is often measured with cross-sectional research studies (Migliavaca et al., 2020). Whilst ‘prevalence’ is closely linked with ‘incidence’ rates, there is an important distinction to make as reports of incident cases indicate the rate of new cases of a condition within a specific time frame (National Institute of Mental Health, 2017). Reviews of prevalence are key in managing service provision and policy (Borges Migliavaca et al., 2020).

Despite the frequency with which fatigue is reported as a concern for those with chronic health conditions, and the far reaching consequences of fatigue (Daniel et al., 2013; Loades & Chalder, 2020), it is often overlooked within interventions (Dittner et al., 2004) and under-acknowledged by clinicians (Lou et al., 2010; Sharpe & Wilks, 2002). It is therefore important to systematically understand the prevalence and nature of fatigue. This review aims to synthesise existing knowledge on the point prevalence of fatigue in children and young people living with chronic health conditions.

Method

The protocol for this review was registered with PROSPERO (CRD42021254251) and reported according to the Preferred Reporting Items for Systematic Reviews and Meta-analysis (PRISMA) guidelines (Page et al., 2021). At the protocol development stage, input from a clinician working within the area of fatigue was sought to maximise the clinical relevance of the review.

Search Strategy

A systematic literature search was conducted using PsycNET, Medline, and Embase databases to identify studies published between January 2000 to July 2021. Search terms were developed in collaboration with a university librarian and based on previous reviews which considered interventions for those with chronic health conditions (Mitchell et al., 2020; Moore et al., 2019; Morey & Loades, 2020) and fatigue (Carroll et al., 2015; Corbett et al., 2019; Spathis et al., 2015). Search terms included both free-text words and Medical Subject Headings (MeSH), with relevant truncations applied to allow variation of search terms. The search strategy focused on four concepts: "child/adolescent", "chronic health condition", "fatigue" and "prevalence" and 'all fields' were searched in each database. Where possible, database filters were applied to limit searches to studies involving humans, with an age range between 0 – 18 years, and published in English. References of included studies were hand searched to identify further relevant papers that may not have been identified within the database searches. The full list of search terms can be found in Appendix B.

Eligibility Criteria

Studies were included if a point prevalence of fatigue for children or adolescents under the age of 18 years was reported, as measured with a validated assessment tool. The age range was chosen following discussion with a clinician working within a paediatric fatigue service and based on the ages of children seen within an NHS paediatric team. When studies included participants over the age of 18 years, we initially planned to contact authors to request data for those only within the 0-18 age bracket. Due to the challenge of obtaining raw data from authors, and following discussion with a clinician, inclusion criteria was adapted to include studies in which participants showed a mean age of 18 years or less.

Participants of studies were also required to have a long term health condition, which persisted for at least three months (Law et al., 2019) and required ongoing

management (NHS, 2020). Traumatic brain injury (TBI) conditions were excluded from this study as previous research (Tomar et al., 2018) has indicated an alternative model of fatigue for those with TBI, based on an increased effort required for everyday tasks due to difficulties with cognitive functioning tasks, such as attention and information processing. TBI conditions were also excluded to avoid replication as a recent meta-analysis (Riccardi & Ciccia, 2021) showed fatigue to be a common symptom for children with TBI. Neurodevelopmental conditions were also excluded based on the varying prognoses (Campbell, 2006) which may affect prevalence of fatigue. In addition, health conditions in which symptoms of fatigue are necessary for diagnosis, such as chronic fatigue syndrome, were excluded. Full eligibility criteria can be found in Table 1.1.

Table 1.1

Inclusion and Exclusion Criteria for Papers

Inclusion criteria	Exclusion criteria
Article is available in English	Article not available in English
The paper is peer-reviewed article, reporting on primary data	Paper has a qualitative design only or is not reporting on primary data e.g., systematic review / meta-analysis
Participants are aged 0 – 18 (or sample mean is ≤18 years)	Participants are not aged between 0 – 18 years old or the sample mean is 19+
Participants have a chronic health condition, with at least 3 months duration.	Participants do not have a chronic health condition – i.e. a condition that lasts at least 3 months, or traumatic brain injury conditions / chronic fatigue syndrome is reported as the primary diagnosis
Fatigue is a reported outcome, measured using a validated assessment tool	Fatigue is not a primary or secondary outcome and is not measured with a validated tool
Study is quantitative or mixed-methods with a point-prevalence report of fatigue	Point prevalence not measured or reported

Study Selection

Following database searches (29-30th July 2021), Covidence online software was used to remove duplicate papers and screen eligible studies. The records were initially screened based on title and abstract, by two of three independent reviewers, with disagreements resolved through discussion. Inter-rated agreement between the reviewers was 'fair' to 'moderate' (McHugh, 2012) at 81 – 95% ($\kappa = 0.26$ and $\kappa = 0.52$). For studies that potentially met inclusion criteria, the full text was screened to confirm eligibility by two independent reviewers, with discrepancies discussed and resolved. If full text was not available, authors were contacted to obtain this. Studies were excluded if the papers were unavailable following two attempts to contact authors.

Data Extraction

Data were independently extracted from eligible studies by two reviewers, with use of a data extraction form which was piloted on three studies. Key information for each study, including the design, country, inclusion criteria, age and sex of participants, fatigue measure, and health condition was extracted. Participant sample size and the number of participants who reported fatigue were recorded to calculate prevalence. If data considered relevant to the review were not reported in the full text, study authors were contacted for further information. Studies were again excluded after two attempts at contacting the author.

Quality Assessment

When we preregistered the review protocol (CRD42021254251), we had intended to use the Joanna Briggs Institute (JBI) Prevalence Critical Appraisal Tool (Munn et al., 2015). Subsequently, piloting the tool showed that this was not suitable for all studies. The methodological quality was instead assessed by two independent reviewers using a modified version of the Newcastle-Ottawa Scale (NOS) for Cross-Sectional Studies (Appendix C) (Stang, 2010; Zhang et al., 2017). This tool includes six questions focusing on the selection process and representativeness of the sample, along with the quality of outcomes. Studies were assessed as either high quality (>3 points) or low quality (≤3 points). Disagreements were resolved through discussion between the two reviewers.

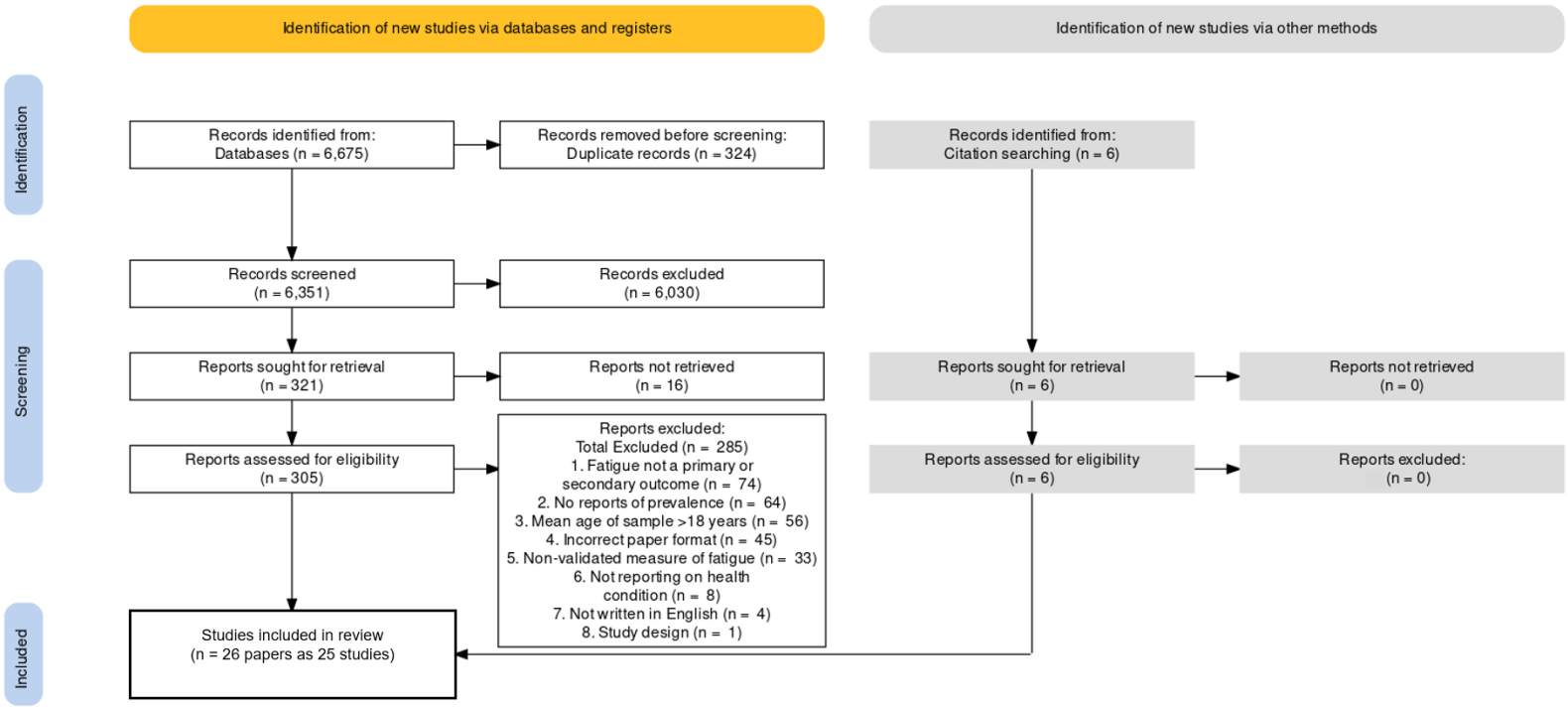
Data Synthesis

We had intended to conduct a meta-analysis, but this was not possible given the heterogeneity of studies which meant that prevalence statistics could not be reliably

pooled together. Instead, a narrative synthesis was used to collate the findings which can offer valuable implications for future research (Arai et al., 2007; Rodgers et al., 2009). The PRISMA reporting guidelines were still followed throughout and the PRISMA flow diagram (Haddaway et al., 2021) (Figure 1.1) shows details of each stage of the review (Page et al., 2021). The Synthesis Without Meta-Analysis guidelines (Campbell et al., 2020), designed to be used alongside PRISMA standards, were also followed.

Figure 1.1

PRISMA Flow Diagram, Including Number of Papers Identified, Screened, and Included



Results

Across the three databases, the search strategy identified a total of 6675 articles, from which 324 duplicates were removed. The initial screening stage excluded 6030 papers. The full texts of the remaining 321 articles were reviewed, with 301 papers excluded largely due to not reporting on fatigue prevalence outcomes and the mean age of participants falling above 18 years. Hand searching the reference list of the 20 included papers led to the identification of a further 6 relevant papers, resulting in a total of 26 papers (25 studies) which met the inclusion criteria.

Study Characteristics

The included research studies focused on fatigue in young people (*n* = 2541 participants) with a broad range of chronic health conditions, with some papers reporting prevalence data for more than one condition. In total, 12 categories of chronic health condition were reported in the included studies, including cancer (28%), autoimmune conditions (19%) and demyelinating conditions, such as MS (19%). The key characteristics of included studies are summarised in Table 1.2. The sample size within studies ranged from 15 to 481, and age of participants ranged from 2 to 22 years, with a mean age of 12.9 (*SD* = 2.9). Overall, the studies included more participants who identified as female (54%) and were conducted predominantly within the USA (32%) and the Netherlands (24%).

Quality Assessment

Thirteen studies were assessed as high quality based on the modified-NOS checklist whilst 12 studies were rated lower quality. The details of this assessment are shown in Table 1.3.

Table 1.2

Key Characteristics of Each Included Paper

Study Information				Health Condition Information		Age	Gender	
Study (Year) Country	Design (Setting)	Aim	Sample Size	Chronic health Condition	Medication / Treatment	Mean, SD, Range	F% (n)	M% (n)
Houghton et al. (2008) Canada	Cross sectional (Outpatients)	Assess fitness, fatigue, and quality of life in young people with systemic lupus erythematosus.	15	Systemic lupus erythematosus	100% taking hydroxychloroquine; 67% taking prednisone; 67% taking at least 1 noncorticosteroid immunosuppressant	16.5 (1.9) 12.8 – 19.6	80% (12)	20% (3)
MacAllister et al. (2009) United States	Cross sectional (Outpatients)	Assess fatigue and quality of life from child and parental perspective, and explore relationship between fatigue, quality of life and clinical factors in MS	51	Multiple Sclerosis	43% treated with Avonex, Betaseron, Copaxone, or Rebif	14.8 (2.21) 9 - 17	64.7% (33)	35.7% (18)
Ketelslegers et al. (2010) The Netherlands	Cross sectional (Community Clinics)	Determine if children with a demyelinating disease of the central nervous system develop fatigue and investigate relationship with depression and health-related quality of life. Comparison between two chronic conditions and a healthy control group	32	Multiple Sclerosis	9.4% treated with Interferon	15.6 (1.47)	60% (6)	40% (4)
Inocente et al. (2014) France	Cross sectional (Community Clinics)	Explore feelings of depression and correlates in children with narcolepsy	88	Narcolepsy	50% never treated 4.5% stopped treatment 45.5% treated for median of 15 months with one or a combination of modafinil, methylphenidate, mzindol, venlafaxine, sodium oxybate	12 (NR) 5 – 17.5	50% (44)	50% (44)
Nunes et al. (2017) Brazil	Cross sectional (Hospital inpatient)	Investigated factors to explain variance of fatigue symptoms	38	Cancer	Chemotherapy, Surgery, Radiotherapy	12.1 (2.9) 8 – 18	34.2% (13)	65.8% (25)
Donnelly et al. (2018) United States	Cohort study (Outpatients)	Assess fatigue, pain, psychological symptoms, disease characteristics and health-related quality of life in young people with childhood-onset	50	Childhood-onset lupus	18% at time point 1 prescribed anti-depressant medication	16.2 (2.5) 11 – 20	84% (42)	16% (8)

		lupus and identify predictors and risk for reduced health-related quality of life.						
Fadhilah & Allenidekania (2019) Indonesia	Cross sectional (Community)	Explore relationship between fatigue and physical activity in children with leukaemia	45	Acute lymphocytic leukaemia	Most received a chemotherapeutic regimen and were in maintenance phase of chemotherapy	NR (NR) 3 – 16	33.3% (15)	66.7% (30)
Lai et al. (2019) United States	Cross sectional (Hospital)	Evaluate symptom burden and associated factors in children with brain tumours	199	Brain tumour	86.5% had received radiation, chemotherapy, or surgical treatment within one year of participating in study	14.1 (3.4) 7 – 22	48.4% (96)	51.6% (103)
Petersen et al. (2019) Germany	Cross sectional (Outpatients)	Investigate fatigue and quality of life in recipients of paediatric liver transplant and compare to healthy controls	100	Paediatric liver transplant recipients	NR	12 (4.5) 2.3 – 18.4	47% (47)	53% (53)
Rogers et al. (2019) United States	Randomised controlled trial (Hospital inpatients)	Primary aim to explore feasibility of a sleep intervention, with secondary aim to investigate relationship between sleep and fatigue	33	Central nervous system cancers	Surgery and 6-weeks of craniospinal radiation	9.5 (3.9) 4 – 19	39.4% (13)	60.6% (20)
Florea et al. (2020) France	Prospective study (Community Clinics)	Explore fatigue, depression symptoms, and quality of life in children with multiple sclerosis and compare with children with acute demyelinating syndromes.	37	Multiple Sclerosis (MS)	NR	15.2 (1.11) 10 – 17	65.4% (17)	34.6% (9)
				Acute Demyelinating Syndromes (ADS)	NR	11.8 (3.11) 10 – 17	54.5% (6)	45.5% (5)
				Full Sample Group (MS and ADS)	NR		63% (23)	37% (14)
Wrightson et al. (2020) Canada	Cohort study (Population)	Characterise fatigue in children with perinatal stroke and investigate relationship with motor performance and corticospinal excitability	45	Perinatal stroke	NR	12 (4) 6 – 18	36% (16)	64% (29)
Goretti et al. (2012) Italy	Cohort study (Community Clinics)	Assess links between fatigue, depression, and cognitive functioning in a paediatric MS population and compare to healthy control group	57	Multiple Sclerosis	87.7% treated with disease modifying drugs	16.6 (2.5) 11 – 20	54% (31)	46% (26)

Jagersma et al. (2013) The Netherlands	Cross sectional (Hospital)	Investigate prevalence of fatigue and assess quality of life in young people with hereditary motor and sensory neuropathy 1A compared to healthy peers and explore effect of fatigue on quality of life	55	Hereditary motor and sensory neuropathy 1A (Charcot-Marie Tooth disease)	NR	15 (2.1) 12 – 18	54.55% (30)	45.45% (25)
Orsey et al. (2013) United States	Prospective study (Hospital)	Investigate relationship between sleep and physical activity in outpatient children with cancer, undergoing treatment	36	Cancer	100% receiving cancer treatment 100% received chemotherapy 30.6% radiation therapy 30.6% steroids	NR (NR) 8 – 18	27.8% (10)	72.2% (26)
Maher et al. (2015) Australia	Cross sectional (Children's Services)	Investigate fatigue and correlates in children with physical disabilities	65	Physical disability including cerebral palsy, brain tumour, spina bifida, Charcot-Marie tooth disease, hereditary spastic paraparesis (62% cerebral palsy)	NR	13.2 (2.7) 8 – 17	46% (30)	54% (35)
Rodrigues-Nunes et al. (2015) United States	Descriptive research design with repeated measures (Hospital)	Investigate sleep patterns and fatigue, and factors associated with fatigue	35	Cancer	NR	12.8 (2.7) 8 – 17	51% (18)	49% (17)
Lai et al. (2016) United States	Cross sectional (Hospital)	Explore prevalence of carnitine deficiency and its relationship with fatigue in young people with cancer who were on-therapy or off-therapy	150	Cancer or Langerhans cell histiocytosis	Receiving treatment or had completed chemotherapy, radiation therapy, stem cell transplant, or surgery	12.75 (3.2) 8 – 17	40.8% (58)	59.2% (84)
Nijhof et al. (2016) The Netherlands	Cross sectional (Outpatients)	Investigate prevalence of fatigue and limitations in adolescents with paediatric rheumatic diseases and compare to healthy control group. Assessed effect of pain and disease activity on severity of fatigue	175	Juvenile idiopathic arthritis and other paediatric rheumatic diseases	45% Methotrexate 36% NSAID 48% DMARD 23% biologic agents 12% corticosteroids	14.5 (2.5) 10 – 18	73.7% (129)	26.3% (46)
Coetzee et al. (2018; 2019) South Africa	Mixed methods (Community Clinics)	Investigate the predictors and correlates of fatigue within adolescents with HIV	134	Human Immunodeficiency Virus (HIV)	Antiretroviral therapy	14.3 (1.9) 11 – 18	58.2% (78)	41.8% (56)

Nap-van der Vlist et al. (2019) The Netherlands	Cross sectional (Outpatients)	Investigate prevalence and extent of fatigue in children and adolescents with chronic health conditions, and compare with two control groups. Further explored impact of fatigue on health-related quality of life	481	Cystic Fibrosis	NR	11.3 (5)	51% (57)	49% (54)
				Autoimmune disease	NR	11.4 (4.4)	65% (179)	35% (98)
				Cancer	NR	7.7 (5)	46% (43)	54% (50)
				Overall Sample Group (all three conditions)	NR	10.7 (4.9) 2 – 18	58% (279)	42% (202)
Vassallo et al. (2020) UK	Cross sectional (Outpatients)	Explore prevalence and severity of fatigue in young people with neurofibromatosis type 1 and unaffected siblings	75	Neurofibromatosis type 1	NR	NR (NR) 2 – 18	53.3% (40)	46.7% (35)
Zimmerman et al. (2020) United States	Cross sectional (Outpatients)	Investigate prevalence and severity of symptoms including fatigue, depression, anxiety, and headaches in children with hydrocephalus	40	Hydrocephalus	Surgically treated hydrocephalus, endoscopic third ventriculostomy (ETV), cerebrospinal fluid shunting	13.5 (NR) 7 – 21	50% (20)	50% (20)
Nap-van der Vlist et al. (2021) The Netherlands	Cross sectional (Outpatients)	Identify biological, psychological, social factors associated with fatigue in children with a chronic disease.	434	Cystic fibrosis	NR	15.3 (NR)	50.7% (36)	49.3% (35)
				Autoimmune disease	NR	14.4 (NR)	63% (165)	37% (97)
				Post-cancer treatment	NR	13.6 (NR)	50.5% (51)	49.5% (50)
				Full Sample Group (all three conditions)	NR	14.5 (NR) 8 – 18	58% (252)	42% (182)
Nijhof et al. (2021) The Netherlands	Cross sectional (Outpatients)	Investigate self and parent report of prevalence of fatigue in children and adolescents with primary immunodeficiency and compare findings with healthy peers. Investigate fatigue effect on health-related quality of life and disease-related factors.	79	Primary Immunodeficiency	NR	10.4 (4.4) 2 – 18	40.5% (32)	59.5% (47)

Note: SD, Standard Deviation; M, male; F, female; NR, Not Reported; NSAID, Non-Steroidal Anti-Inflammatory Drugs; DMARD, Disease-Modifying Antirheumatic Drugs

Table 1.3

Results of Quality Assessment Using a Modified-NOS for Cross-Sectional Studies

Study	Representativeness of the Sample (One Point)	Sample Size (One Point)	Nonrespondents (One Point)	Ascertainment of Exposure (One Point)	Assessment of Outcome (One Point)	Descriptive Statistics Reporting (One Point)	Score	Descriptive
Houghton et al. (2008)	0	0	0	1	1	1	3	Low quality
MacAllister et al. (2009)	0	0	0	1	1	0	2	Low quality
Ketelslegers et al. (2010)	1	0	0	1	1	0	3	Low quality
Inocente et al. (2014)	1	0	0	1	1	0	3	Low quality
Nunes et al. (2017)	0	0	0	1	1	1	3	Low quality
Donnelly et al. (2018)	0	0	0	1	1	1	3	Low quality
Fadhilah & Allenidekania (2019)	0	0	0	0	0	0	0	Low quality
Lai et al. (2019)	1	0	0	1	1	0	3	Low quality
Petersen et al. (2019)	0	0	0	1	1	0	2	Low quality
Rogers et al. (2019)	0	0	0	1	1	0	2	Low quality
Florea et al. (2020)	1	0	0	1	0	0	2	Low quality
Wrightson et al. (2020)	1	0	0	1	1	0	3	Low quality
Goretti et al. (2012)	1	0	0	1	1	1	4	High quality
Jagersma et al. (2013)	1	0	1	1	1	0	4	High quality
Orsey et al. (2013)	1	0	0	1	1	1	4	High quality
Maher et al. (2015)	1	0	0	1	1	1	4	High quality
Rodrigues-Nunes et al. (2015)	1	1	0	1	1	1	5	High quality
Lai et al. (2016)	1	0	0	1	1	1	4	High quality
Nijhof et al. (2016)	1	0	0	1	1	1	4	High quality
Coetzee et al. (2018; 2019)	1	0	0	1	1	1	4	High quality
Nap-Van Der Vlist et al. (2019)	1	1	1	1	1	1	6	High quality
Vassallo et al. (2020)	0	0	1	1	1	1	4	High quality
Zimmerman et al. (2020)	0	1	0	1	1	1	4	High quality
Nap-Van Der Vlist et al. (2021)	1	0	1	1	1	1	5	High quality
Nijhof et al. (2021)	0	1	0	1	1	1	4	High quality

NOS = Newcastle-Ottawa Scale; Ratings: High quality 4-6; Low quality 0-3

Fatigue

Measurement

Across the 25 studies, ten measures of fatigue were used, with one study (Lai et al., 2016) reporting prevalence based on two different assessment tools. The Paediatric Quality of Life Inventory – Multidimensional Fatigue Scale (PedsQL-MFS) (Varni et al., 2002) was the most commonly used assessment tool and was included in almost half of the studies (*n*=12). All 25 studies asked children or young people themselves to self-rate their levels of fatigue. Additionally, nine studies included a version of parental report; four of which were a parent-proxy rating for children under the age of eight, a further three papers included a parent-child dyad rating, and two recent studies (Petersen et al., 2019; Vassallo et al., 2020) included both proxy and dyad ratings.

Prevalence

Overall, within the included studies (*n*=25), 52 estimates of the prevalence of fatigue were reported with a multitude of heterogeneity based on a diverse combination of groupings of health conditions, age range, severity of fatigue and self or multi-raters. For example, some papers report prevalence based on level of fatigue, with calculations based on participants who meet the threshold for ‘severe’ fatigue only, whilst other papers report total ‘abnormal’ fatigue which included those scoring within the mild and moderate ranges on validated assessment tools. Other papers combined multiple health conditions into their grouping of fatigue prevalence whilst some authors calculated the prevalence of fatigue for specific conditions. Additionally, one paper (Rogers et al., 2019) divided prevalence of fatigue into age ranges and reported this separately for child age range and adolescents.

Amongst the nine studies that included a form of parental report, further variation was noted; in some papers, parental-proxy reports were gathered for children under the age of eight and prevalence scores were reported within the same score as children who were able to complete an assessment of fatigue themselves, others still created a separate parent-rated grouping for prevalence estimates which included reports from both parent-proxy and parent-dyad measures. This adds further complexity and heterogeneity to the data and so prevalence rates cannot be meaningfully pooled into a meta-analysis.

Prevalence estimates based on self-reported levels of fatigue ranged from 5% (Ketelslegers et al., 2010) to 100% (Fadhilah & Allenidekania, 2019) – although both of these studies were rated as low in quality. When taking only high-quality studies into

account, self-report prevalence ranged from 14% for total identifiable fatigue (Goretti et al., 2012) to 72% (Lai et al., 2016). As can be seen in Table 1.4, self-ratings of fatigue were notably different to parental reports, with parents in all but one study (Lai et al., 2019) reporting a higher prevalence of fatigue than children and young people reported themselves. Where parent reports were presented separately from self-ratings (6 studies), parental prevalence estimates ranged from 38% (Lai et al., 2019) to 69% (Vassallo et al., 2020) for total identifiable levels of fatigue. In the same studies, prevalence ratings based on self-report from children and young people ranged between 14% (Goretti et al., 2012) and 57% (MacAllister et al., 2009). On average, prevalence based on parental reports was 26% higher than prevalence based on self-reports of fatigue, and the ratings from parents of children with primary immunodeficiency showed the largest discrepancy (43%).

In general, studies that reported higher prevalence were rated as lower in quality; seven studies reported fatigue to be present in over 60% of participants and only one of these studies (Lai et al., 2016) was assessed to be of high quality. The highest prevalence ratings were typically reported in studies with paediatric cancer patients; of the nine studies within this population, six reported fatigue to be present in over 50% of their sample. Outside of this population, prevalence was reported to be highest and most consistent among those with lupus-conditions; two studies (Donnelly et al., 2018; Houghton et al., 2008) explored fatigue in lupus and reported similar prevalence estimates (66 – 67%).

Interestingly, whilst lupus is considered an autoimmune condition, the four other studies which investigated autoimmune disorders within the paediatric community reported much lower prevalence estimates of fatigue. Of the studies (3) that reported separate prevalence ratings for an autoimmune condition, such as HIV and JIA, prevalence estimates ranged from 20 to 25% (Coetzee et al., 2018; Coetzee et al., 2019; Nap-van der Vlist et al., 2021; Nijhof et al., 2016).

Eight of the included studies, six of which were assessed to be high quality, compared fatigue in those with a chronic health condition to either an actively recruited healthy control group (*n*=3) and/or previously published data for healthy controls (*n*=6). The majority of these studies (*n*=7) made comparisons based on the level or severity of self-reported fatigue, with five studies demonstrating significantly higher levels of fatigue across chronic health conditions. In addition, some of the studies (*n*=3) drew comparisons based on prevalence of fatigue, with all three studies demonstrating that higher numbers of paediatric patients scored above clinical thresholds for fatigue, compared to healthy peers.

The majority of studies were conducted in the USA (*n*=8) and the Netherlands (*n*=6). Across conditions, studies conducted in the USA reported prevalence ranging from 35 – 71%. In contrast, studies conducted in the Netherlands reported lower prevalence from 16 – 37%.

Correlates

The majority of included studies (*n*=22) reported on factors associated with the presence or severity of fatigue and highlighted biopsychosocial components. Seven studies found an association between fatigue and quality of life, with one (Donnelly et al., 2018) longitudinally highlighting that for young people with lupus higher levels of fatigue, alongside depression, predicted poorer quality of life at six month follow up. Overall, symptoms of depression were shown to have weak to strong correlations with fatigue in five studies, whilst anxiety was explored in only two studies (Coetzee et al., 2018; Zimmerman et al., 2020) and was not reported to have a significant correlation. Difficulties and disturbances with sleep were significantly associated with fatigue in five studies, most commonly showing a weak association, and physical functioning was also reported as a (small to moderate) correlate with fatigue in four of the studies. Experiences at school, including pressure (one study) and absence (two studies), and social functioning (one study) also showed weak but significant correlations with fatigue.

In one study (Rogers et al., 2019), the presence of fatigue was compared across children and adolescents with cancer. This demonstrated a higher prevalence of fatigue in adolescents compared to self-report and parental-proxy report for children under the age of 12. Similarly, Nap-van der Vlist and colleagues (2021) reported higher levels of fatigue in the older paediatric age group but did not report on how prevalence differed across age ranges. In total, four studies demonstrated weak but significant relationships between fatigue and older age, whilst two studies showed no association in this area (Maher et al., 2015; Wrightson et al., 2020).

Other studies also identified relationships between fatigue and demographics across a range of conditions, including female sex (three studies) and low social economic status (one study). One study with young people with JIA (Nijhof et al., 2016) demonstrated a link between levels of fatigue and pain, however another five studies, all within the autoimmune population, consistently highlighted no association between fatigue and characteristics of the condition, such as disease activity and treatment.

Table 1.4

Fatigue Prevalence Across Included Studies

Study (Year)	Chronic health Condition	Measure of Fatigue	Rater	Prevalence of fatigue (numerator/ denominator)	Mean Fatigue Score (SD)	Findings
Houghton et al. (2008)	Systemic lupus erythematosus	Kids Fatigue Severity Scale	Self-report, ages 12-19	66.7% (10/15)	3.8 (1.2)	Fatigue is a major symptom in young people with systemic lupus erythematosus. No significant relationship was found between fatigue and aerobic fitness or quality of life.
MacAllister et al. (2009)	Multiple Sclerosis	PedsQL MFS	Self-report, ages 7-19	Mild 24% (12/49) Severe 32% (16/49) Total 57.1% (28/49)	NR (NR)	Self-report and parent-reports of child fatigue were significantly higher than healthy control samples.
			Parent dyad, ages 7-19	Mild 12% (6/47) Severe 51% (24/47) Total 63.8% (30/47)	NR (NR)	
Ketelslegers et al. (2010)	Multiple Sclerosis	Checklist Individual Strength	Self-report, ages 11-17	40% (4/10)	NR (NR)	Children with multiple sclerosis were more likely to experience severe fatigue than healthy peers. Fatigue is less likely in children with monophasic inflammatory demyelinating disease. Possible relationship between fatigue and depression and health-related quality of life
	Monophasic conditions			5% (1/22)	NR (NR)	
	Full Sample Group (MS and Monophasic conditions)			16% (5/32)	NR (NR)	
Inocente et al. (2014)	Narcolepsy	Chalder Fatigue Scale	Self-report, ages 5-17	26.7% (23/86)	8 (NR)	A quarter of participants (mostly girls over the age of 10 years) experienced high levels of depression, which were associated with fatigue. Fatigue also correlated with excessive daytime sleepiness, insomnia, and hyperactivity.
Nunes et al. (2017)	Cancer	PedsQL MFS	Self-report, ages 8-18	66.7% (25/38)	63.8 (18.5)	Children and adolescents with cancer had difficulties with three dimensions of fatigue (general, sleep/rest, and cognitive). Significant correlation was found between fatigue and reduced health-related quality of life.

Donnelly et al. (2018)	Childhood-onset lupus	PedsQL MFS	Self-report, ages 11-20	66% (33/50)	58.6 (18.9)	Reduced health-related quality of life at follow up was predicted by higher levels of fatigue and depression symptoms at initial visit.
Fadhilah & Allenidekania (2019)	Acute lymphocytic leukaemia	Allen's Fatigue in Childhood Cancer Scale	Self-report, ages 3-16	100% (45/45)	Less physically active 10.65 (NR) Physically active 6.27 (NR)	Higher levels of physical activity are associated with lower fatigue in children with leukaemia whilst at home.
Lai et al. (2019)	Brain tumour	PROMIS	Self-report, ages 7-22 Parent dyad, ages 7-22	39% (78/199) 38.3% (76/199)	44.6 (13) NR (NR)	Participants reported less anxiety, depression, and fatigue than the norming sample. Fatigue, anxiety, depression, mobility, peer relationships, and cognition were correlated with symptom distress
Petersen et al. (2019)	Recipients of liver transplant	PedsQL MFS	Self-report, ages 8-18 Parent proxy, ages 2-7 parent dyad, ages 2-18	37% (26/71) 57% (57/100)	74 (15.29) NR (NR)	Children and parents both reported significantly more fatigue than healthy peers. General and cognitive fatigue were significant predictors of health-related quality of life in self report and parent proxy report.
Rogers et al. (2019)	Central nervous system cancers	Fatigue Scale C/A/Parent Fatigue Scale Child / Parent Fatigue Scale Adolescent	Self-report, ages 7-19 Parent proxy, ages 4-6 Self-report, ages 7-12 Parent proxy, ages 4-6 Self-report, ages 13-19	60% (20/33) 53.3% (13/24) 77.8% (7/9)	NR (NR) NR (NR) NR (NR)	Higher levels of fatigue were associated with higher levels of night-time activity and lower percentage of sleep in adolescents but longer sleep episodes in children - as measured by an actigraph.
Florea et al. (2020)	Multiple Sclerosis Acute Demyelinating Syndromes	Fatigue Severity Scale	Self-report, ages 10-17	43% (10/23) 63% (5/8)	NR (NR) NR (NR)	Fatigue is a key symptom in young people with MS and those with ADS, with children with ADS reporting fatigue more frequently.
Wrightson et al. (2020)	Perinatal stroke	PedsQL-CP	Self-report, ages 8-18 Parent proxy, ages 6-8	46.6% (21/45)	75 (NR)	Almost half reported experiences of fatigue in the previous month, showing that fatigue is a common experience for children with hemiparesis with perinatal stroke. No relationship found between fatigue and age, type of stroke, or gender.

Goretti et al. (2012)	Multiple Sclerosis	PedsQL MFS	Self-report, ages 11-20	Mild 9% (5/57) Severe 5% (3/57) Total 14% (8/57)	78.5 (19.8)	A sizeable proportion of children with MS are affected by fatigue and fatigue was significantly associated with higher levels of depressive symptoms. Differences between self- and parent-report fatigue levels is noted.
			Parent dyad, age 11-20	Mild 17.5% (10/57) Severe 21% (12/57) Total 39% (22/57)	74.3 (17.7)	
Jagersma et al. (2013)	Hereditary motor and sensory neuropathy 1A (Charcot-Marie Tooth disease)	Checklist Individual Strength	Self-report, ages 12-18	24% (13/55)	NR (NR)	There is a high prevalence of severe fatigue and reduced quality of life reported in children with hereditary motor and sensory neuropathy 1A, compared with healthy peers.
Orsey et al. (2013)	Cancer	Fatigue Scale C/A (24h)	Self-report, ages 8-18	58% (21/36)	Child 17 (NR) Adolescent 28 (NR)	Morning mood negatively correlated with a 7-day rating of fatigue, and sleep quality correlated with both 24 hour and 7-day ratings of fatigue. Whilst fatigue correlates with sleep quality, fatigue on its own was not significantly linked with sleep disturbance.
		Fatigue Scale C/A (7 day)	Self-report, ages 8-18	56% (20/36)	Child 19 (NR) Adolescent 26 (NR)	
Maher et al. (2015)	Physical disability including cerebral palsy, brain tumour, spina bifida, Charcot-Marie tooth disease, hereditary spastic paraparesis (62% cerebral palsy)	PedsQL MFS	Self-report, ages 8-17	37% (24/65)	65.5 (14.4)	Fatigue was associated with female gender, low socio-economic status, and being physically inactive. No associations found between fatigue and age, weight, or functional impairment. Those with physical disabilities reported high levels of fatigue compared to other paediatric health populations, with reported fatigue comparable to levels reported in paediatric cancer patients.
Rodrigues-Nunes et al. (2015)	Cancer	PedsQL MFS	Self-report, ages 8-17	54% (19/35)	76.2 (15.4)	Over half of the participants reported difficulties with fatigue at home. Adolescents reported more problems with fatigue than children, and females reported more fatigue than males. Fatigue also varied by cancer diagnosis, with those with sarcoma reporting more fatigue than those with other cancer diagnoses.
Lai et al. (2016)	Cancer or Langerhans cell histiocytosis	pedsFACIT-F	Self-report, ages 8-17	56.3% (80/142)	NR (NR)	No significant relationship between fatigue and carnitine levels were found. Those who were currently undergoing treatment reported more severe fatigue than those who had completed cancer treatment.
		PedsQL MFS		71.8% (102/142)	NR (NR)	

Nijhof et al. (2016)	Juvenile idiopathic arthritis and other paediatric rheumatic diseases	Checklist Individual Strength	Self-report, ages 10-18	25.1% (44/175)	28.2 (13.8)	Severe fatigue was associated with increased absence from school and reduced physical functioning. Association found between fatigue and pain, but not with disease activity.
Coetzee et al. (2018; 2019)	HIV	Chalder Fatigue Scale	Self-report, ages 11-18	24.6% (33/134)	14.89 (3.83)	Fatigue was more likely to be reported by older adolescents, those with more difficulties with sleep, and those with higher levels of depression. Depression alone did not account for all fatigue experienced by those with HIV.
Nap-Van Der Vlist et al. (2019)	Cystic Fibrosis	PedsQL MFS	Self-report, ages 8-18 and Parent proxy, ages 2-7	NR (NR/NR)	72 (18.6)	There is a high prevalence of fatigue in children and adolescents with chronic health conditions. Fatigue is likely across disease, age and gender and impacts quality of life.
	Autoimmune disease			NR (NR/NR)	74.5 (18.3)	
	Cancer			NR (NR/NR)	75.3 (18.6)	
	Overall Sample Group (all three conditions)			37% (178/481)	NR (NR)	
Vassallo et al. (2020)	Neurofibromatosis type 1	PedsQL MFS	Self-report, ages 5-18	34% (18/53)	55 (19)	Perceived fatigue affects children with NF1 more than healthy children without NF1.
			Parent proxy, ages 2-4 Parent dyad, ages 5-18	69% (49/71)	53.5 (20.1)	
Zimmerman et al. (2020)	Hydrocephalus	PROMIS	Self-report, ages 7-21	Mild 7.5% (3/40) Moderate 10% (4/40) Severe 15% (6/40) Total 35% (13/40)	45.1 (16.4)	No significant relationship between fatigue, anxiety, depression, or demographics. Mean scores of fatigue, anxiety and depression were similar to those found in paediatric patients with other chronic health conditions, such as cancer, kidney disease, and asthma.
Nap-Van Der Vlist et al. (2021)	Cystic fibrosis	PedsQL MFS	Self-report, ages 8-18	21.1% (15/71)	75 (NR)	Fatigue in children with chronic health conditions is multidimensional and associated with bio-psycho-social factors. Higher levels of fatigue was associated with reduced physical functioning, increased depressive symptoms, more pressure within school, poorer social functioning and older age.
	Autoimmune disease			20.6% (54/262)	75 (NR)	
	Post-cancer treatment			26.7% (27/101)	75 (NR)	
	Full Sample Group (all three conditions)			21.8% (95/434)	75 (NR)	
Nijhof et al. (2021)	Primary Immunodeficiency	PedsQL MFS	Self-report, ages 8-18	18.9% (10/53)	69.7 (16.9)	Fatigue is prevalent in young people with PID. Severe fatigue was linked with increased absence from school and reduced quality of life. It may be beneficial for interventions to focus on fatigue to increase school engagement and quality of life.
			Parent proxy, ages 2-7	62.5% (15/24)	61.8 (16.6)	

Note. PedsQL MFS, Paediatric Quality of Life Inventory Multidimensional Fatigue Scale; pedsFACIT-F, Paediatric Functional Assessment of Chronic illness therapy-fatigue; PedsQL-CP, Paediatric Quality of Life Inventory Version 3.0 Cerebral Palsy (fatigue subscale); Fatigue Scale C/A, Fatigue Scale Child/Adolescent; PROMIS, Patient Reported Outcome Measurement Information System; NR, not reported; HIV, Human Immunodeficiency Virus; NF1, Neurofibromatosis type 1; MS, Multiple Sclerosis; ADS, Acute Demyelinating Syndrome; PID, Primary Immunodeficiency

Discussion

Our review of what is known about the point prevalence of fatigue in children and young people with chronic health conditions captured a total of 26 papers (25 studies) in which fatigue was measured with a variety of assessment tools, across a range of conditions, and the prevalence reported in heterogenous ways. Overall, 52 rates of prevalence were reported within groups of different health conditions, or reported separately based on specific conditions, severity of fatigue, self-report, and parental reports. The prevalence of fatigue varied widely across studies; including studies assessed as lower quality, prevalence estimates ranged from 5 to 100%, whilst higher quality studies only reported a reduced prevalence range from 14 to 72% based on total identifiable levels of fatigue. Discrepancies were also noted in studies that investigated similar populations, with the prevalence of fatigue in those with MS, for example, ranging from 14 to 57%.

Even within the same study, vast discrepancies were noted between child reported fatigue and parental reports. The studies showed that parents tended to report a higher prevalence of fatigue than children and young people reported in themselves; prevalence based on parental report ranged from 38% to almost 70%, whilst the same studies showed that the prevalence of self-reported fatigue ranged from 14 – 57%. These findings are consistent with a previous systematic review (Carroll et al., 2015) which investigated fatigue in children and adolescents with MS and noted similar discrepancies between child and parent reports. Goretti and colleagues (2012) posit that such discrepancies could be due to under-reporting by children and young people, over-estimating from parents, or a combination of the two. Previous research has demonstrated a relationship between family functioning and fatigue in children with life-threatening health conditions (Huang et al., 2013) which may be reflected in parental responses to fatigue measures. Furthermore, inconsistencies between child and parental reports has been demonstrated in other areas, such as health-related quality of life, with researchers suggesting that wellbeing of parents may impact their report of child symptoms (Eiser & Varni, 2013). It is also possible that child developmental stage affects insight and metacognitive reflective abilities required for self-reporting fatigue, as has been found in health-related quality of life (Conijn et al., 2020), which may further create discrepancies between parent and self-report. This highlights the difficulties in measuring fatigue, both in research and clinically, particularly in young children or those with developmental needs, who may be unable to complete measures themselves. Where possible, it may be beneficial to include multiple

perspectives to gather a more thorough sense of how fatigue affects children and young people.

Each of the included studies investigated fatigue in children and young people with different chronic health conditions, at separate stages of condition, and who were accessing different medical regimes. Given that fatigue is a subjective and multi-faceted experience which can be related to biological, psychological, and social factors (Loades & Chalder, 2020; Surawy et al., 1995), as well as treatment regime (Al Maqbali et al., 2021), it is perhaps not unexpected that this review found such a vast range of prevalence reports across studies.

Across the studies, ten measures of fatigue were included, with child and parent versions of the PedsQL-MFS (Varni et al., 2002) used most frequently. This measure was one of only two recommended assessments for children and young people with chronic health conditions in a previous systematic review (Crichton et al., 2015) which showed it to be a valid measure across conditions and useful for the assessment of younger children, from the age of 2, as well as adolescents up to 18. Of the studies that investigated fatigue in the younger age group, all but one included the PedsQL-MFS. The authors also recommended the Fatigue Scale (Hinds et al., 2010) specifically for a paediatric cancer population. Whilst this assessment was used less often in the included studies, this may be a result of many of the paediatric cancer studies opting to use the PedsQL-MFS as it allows more comparability with other conditions. Overall, it is promising that over half of the studies incorporated one of the two recommended measures when assessing fatigue.

Despite the discrepancies in measures and prevalence reports, both within and between studies, some commonalities were identified. For example, several studies investigated the presence or severity of fatigue in young people with a chronic health condition and drew comparisons with a sample of healthy peers. In all of these studies, a higher prevalence of fatigue was found in those with a chronic health condition, and a further five papers demonstrated an increased severity of fatigue in the chronic health group. This indicates that at least some level of fatigue is common across conditions and affects those with health needs more often than healthy peers. This is consistent with findings from a population-based adult cohort study in which a higher prevalence of participants with one or more chronic conditions reported severe and chronic fatigue compared to those without health conditions (Goërtz et al., 2021).

Studies investigating fatigue in the paediatric cancer population were most common, and in general showed fatigue to be highly prevalent, with six of the nine cancer studies reporting a prevalence of above 50%. Fatigue in young people with cancer fell below this rate when prevalence was reported in combination with different health conditions, and in one study which focused only on young people with brain tumours. This reported prevalence range in children and young people with cancer appears to be higher than has been found in a recent review (Al Maqbali et al., 2021) which explored prevalence of fatigue in patients with cancer aged 15 and older. Similarly though, the recent review reported significant heterogeneity between studies with variations in prevalence based on treatment and diagnosis.

The majority of studies explored the relationship between fatigue and biological, psychological or social aspects of life. Associations between fatigue and quality of life, depression and sleep were most commonly demonstrated across conditions, with one longitudinal study (Donnelly et al., 2018) showing that fatigue and depression were associated with poorer quality of life six months later. Similar findings have been demonstrated in a recent review which investigated the correlates of fatigue in older adults with chronic health conditions (Torossian & Jacelon, 2021) and suggested a common underlying pathway of fatigue and associated qualities across health conditions (Hardy & Studenski, 2010). These correlates link to the cognitive behavioural model of fatigue (Surawy et al., 1995) which highlights the role of emotional and behavioural factors in maintaining symptoms of fatigue.

A recent systematic review (Al Maqbali et al., 2021) with those with cancer, showed that fatigue appeared to vary by date, with reported prevalence decreasing by over 20% in recent years. We did not see this pattern in our review. However the prevalence of fatigue did appear to differ between countries, with studies in the US reporting a point prevalence ranging from 35 to 71% whilst prevalence within studies conducted in the Netherlands ranged from 16 to 37%. Whilst this is inconsistent with previous research (De Kleijn et al., 2009) which has shown similar prevalance ratings between adults with sarcoidosis in the US and the Netherlands, it is possible that reports of fatigue may vary across cultures due to assessment of the concept of fatigue or medical regime (Karasz & Mckinley, 2007). It may be helpful for future research to explore this further.

Strengths and Limitations

The exclusion of gray literature and restrictions of language and date, across only three databases, may have excluded some studies relevant to this review. Given the heterogeneity within the included studies, it was not possible to conduct meta-analysis, which further limits the generalisability of the findings. The inclusion, however, of a wide range of chronic health conditions, across different countries, and in populations with a mean age of 18 years or under, strengthens the conclusion that at least some level of fatigue appears to be present across chronic health conditions within children and young people.

Clinical Implications

Previous research suggests that fatigue is often overlooked within healthcare (Dittner et al., 2004), however this review indicates that fatigue is common and can be severe in children and young people with chronic health conditions and should be routinely assessed within clinical settings, particularly due to the potential impact on quality of life, mood, and school attendance. Such factors may also be important to consider as part of clinical formulations, based on the cognitive behavioural model of fatigue (Surawy et al., 1995). Discrepancies of symptoms may be common between children and parents, therefore gathering information from multiple sources can inform clinical discussions and allow exploration of family dynamics and impact on family functioning (Vetter et al., 2012). Promising psychological interventions for adolescents with fatigue, including the use of psychoeducation and cognitive behavioural therapy, have been identified in a recent systematic review (Higson-Sweeney et al., Submitted) which could be beneficial for children and young people with chronic health conditions. At a service level, the NHS Long Term Plan (NHS, 2019) sets out guidance for joined up care between services; this review indicates that collaborative working between health settings and fatigue services may be helpful for children and young people with chronic health conditions.

Future Research

Whilst the majority of studies included an investigation of correlates of fatigue, this was only explored in this review alongside prevalence reports. Future research should systematically investigate the relationship between fatigue and biological, psychological, and social factors in children and young people with chronic health conditions. Understanding this will allow for potential transdiagnostic interventions within healthcare services. In addition, this review noted discrepancies between child and parental reports of fatigue; further understanding of the variation in symptom reporting may benefit

services in gathering information and utilising outcome measures. It is also important to ensure that young people's voices are heard within the evidence-base, mixed-methods studies will be important to capture the perspectives of children and young people regarding fatigue and its impact on their lives. In future, systematic reviews may look at synthesising mixed-methods or qualitative studies to gain a more thorough understanding of the nature of fatigue within this population.

Conclusion

In conclusion, whilst there is a discrepancy in the estimated prevalence of fatigue within children and young people across chronic health conditions, the review shows that some level of fatigue is present across conditions and tends to be higher in those with chronic health conditions compared to healthy peers. A combination of self-report and parental report may be required in services to understand how fatigue affects young people and routine screening for fatigue may inform psychological support which could lead to improvements in other areas of life, such as quality of life and mood. Further studies should explore such correlates in more detail.

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Service-Related Project

Evaluation of a mindful life group: A service-related project

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Proposed Journal: Mindfulness (Appendix D)
This journal publishes manuscripts regarding the research, theory, and clinical practice of mindfulness, and accepts papers under 45 pages.

A lay summary is available in Appendix E.

Evaluation of a mindful life group: A service-related project

Abstract

Objectives: This service improvement project evaluated a twelve-week mindfulness group facilitated with people with a range of difficulties across adult and later life secondary mental health teams. The study aimed to evaluate the impact of the group on levels of mindfulness and psychological distress. **Methods:** Participants were those who completed the group (*n*=70) and completed two standardised self-report measures of distress (*n*=38) and mindfulness (*n*=40) which were completed at the beginning and end of the intervention, over a five-year period and with 14 cohorts of the group. **Results:** There was a significant increase in overall levels of mindfulness over the course of the intervention, with a medium effect size, as well as within three specific components of mindfulness - non-reacting, observing and non-judging. A small but non-significant decrease in psychological distress was noted from the start to the end of the group and not reacting to inner experiences is presented as a key mindfulness skill in supporting the reduction of distress. **Conclusions:** Mindfulness-based skills increased over the twelve-week intervention and small reductions in distress were noted. Improvements in psychological distress may be further supported with additional practices which concentrate on developing a non-reactive stance. The implications for the service, alongside recommendations for future-research, are discussed.

Introduction

Mindfulness

Research into the use of mindfulness as a clinical intervention has grown rapidly over recent decades (Goldberg et al., 2017), although studies often note that the use of varied definitions of 'mindfulness' can affect the clarity of the research (Coffey et al., 2010; Gu et al., 2015). Common definitions of mindfulness often focus on the development of an accepting, non-judgemental and purposeful awareness of the present experience (Gu et al., 2015; Ludwig & Kabat-Zinn, 2008), with Shapiro and colleagues (2018) identifying intention, attention, and attitude as three key elements to this process.

Whilst originally based on Buddhist practice (Lazaridou & Pentaris, 2016), today the use of mindfulness transcends religion (Williams & Kabat-Zinn, 2011) and it has now become an effective and feasible psychological intervention (Radford et al., 2012). It is often used in collaboration with a range of psychological therapies to improve mental health and wellbeing (Cayoun, 2011), with elements of mindfulness practice being integrated within third wave Cognitive Behaviour Therapy approaches (Harrington & Pickles, 2009) such as dialectical behavioural therapy (Linehan, 1993) and acceptance and commitment therapy (Hayes & Wilson, 1994). Structured mindfulness-based interventions, such as Mindfulness-Based Cognitive Therapy (MBCT) (Segal et al., 2002) and Mindfulness-Based Stress Reduction (MBSR) (Kabat-Zinn & Hanh, 2009), have also evolved and grown in popularity in their own right (Hofmann et al., 2010). These interventions are both delivered as manualised group programs, across eight-weeks, and include a selection of practices such as mindful movement, mindfulness of day to day activities, and breath or body-focused practice (Felder et al., 2012).

MBCT interventions, specifically, are underpinned by a cognitive theory of depression, whereby low mood is associated with negative thought patterns (Kuyken et al., 2010), as well as the theory that developing mindfulness skills and practice can cultivate an accepting and non-reactive stance, whereby cognitive and emotional reactions to stress are reduced (Segal et al., 2002; Sipe & Eisendrath, 2012). This model is supported by Gu and colleagues (2015) in a systematic review of the mechanisms behind the effectiveness of MBCT and MBSR interventions. In addition to reactivity being a strong mediator, the review also demonstrated that mindfulness and repetitive negative thinking processes, such as rumination and worry, are significant mediators of psychological outcomes. This suggests mindfulness interventions work by helping one to develop a metacognitive awareness of, and ability to disconnect from, negative thinking

cycles and reactive states (Gu et al., 2015; Sipe & Eisendrath, 2012). This in turn supports the regulation of emotions and alternative coping skills (Coffey & Hartman, 2008).

Research has shown mindfulness-based interventions to be effective at reducing psychological distress in both clinical (Compen et al., 2018; Proeve et al., 2018) and non-clinical populations (Martín-Asuero, 2010; Virgili, 2015). Furthermore, randomised controlled trials have demonstrated that mindfulness-based interventions support symptom reduction in specific mental health presentations, such as generalised anxiety disorder (Hoge et al., 2013), recurrent depression (Godfrin & Van Heeringen, 2010), emotionally unstable personality disorder (Elices et al., 2016) and psychosis (Chien & Lee, 2013). Beyond a reduction in symptoms, researchers acknowledge that mindfulness interventions can be associated with other outcomes, including increased mindfulness, quality of life, cognitive flexibility, improvements in pain and reduced risk of relapse for those with a history of depression (Hazlett-Stevens, 2018; Kuyken et al., 2016; Reiner et al., 2013; Zou et al., 2020).

Whilst much of the research focuses on diagnosis-specific groups, the value of mindfulness-based interventions within heterogenous outpatient samples has also been evidenced (Ree & Craigie, 2007). Radford and colleagues (2012) demonstrate that group-based mindfulness interventions can be usefully applied within clinical settings with a range of mental health presentations. This pilot study reported improvements in a range of outcomes, including depression, rumination, anxiety, and wellbeing – which were maintained at six-month follow up. Whilst research also tends to focus on working age adults, investigations around efficacy of mindfulness interventions have also been conducted across the lifespan (Broderick & Jennings, 2012; Flook et al., 2015; Gallegos et al., 2013), with findings suggesting that it can be feasible and acceptable with older adults (Geiger et al., 2016) and may moderate effects of stress on mental health (de Frias & Whyne, 2015). A recent review, however, concludes that whilst preliminary evidence into mindfulness-based treatment of anxiety and depression in older adults appears positive, further research is needed (Thomas et al., 2020).

The Service

Between 2015 and 2020, an NHS secondary mental health team in the UK has facilitated a 12-week ‘Mindful Life’ group for clients across ages and diagnoses. This group is based on the eight-week manualised MBCT group (Segal et al., 2012) and has been adapted to become more suitable for clients with current or chronic mental health difficulties. In addition to the extended course length, the group encompasses a longer discussion time with fewer practices ranging from five to 10 minutes in length. A further

adaptation has been made to the length of sessions, as each session is briefer at 1.5 hours rather than the traditional two hours of MBCT.

The group is run by two co-facilitators, a Clinical Psychologist and Clinical Nurse Specialist. The group runs three times a year, with each cohort consisting of between five to 10 attendees. Those attending can be referred by, and care-coordinated under, four different services which include an adult psychological therapies service, a later life team, an early intervention (EI) service, and recovery team. This means that those attending the group present with a diverse range of diagnoses and difficulties. Such presentations include anxiety, depression, psychosis, mild cognitive impairment, Alzheimer's Disease, physical health conditions, chronic pain, or alternatively attendees may have a carer role. Before starting the group, facilitators meet individually with referred clients to conduct risk assessment, consider group suitability, and assess motivation and commitment for attending the group. The group format starts with encouraging discussion and offering psychoeducation around what mindfulness is and rationale for practice, this then leads on to various themes, such as mindfulness of body, pain, action, thoughts, and emotions. Clients are encouraged to attend all twelve sessions as each week covers a different theme.

Previous research within the service (Armstrong, 2019) has explored qualitative responses to the 'Mindful Life' group (n = 8); this found that many described the intervention as beneficial – however it is not known whether this finding is supported by the quantitative outcome measures and attrition rates of those who have been invited to the group across the five years. The present evaluation provided an opportunity to explore whether there are any changes that could be implemented to increase the efficacy of the group for service users (de Lichtenberg & London, 2008). This research also sought to build on the preliminary evidence on the use of mindfulness for heterogenous outpatient groups (Radford et al., 2012).

Aims

This research forms a service development project and aimed to provide an evaluation of quantitative measures collected as part of the 12-week Mindful Life group, with a view of offering recommendations for how the group may be adapted in the future. Overall, the study aimed to investigate the impact of the mindfulness group intervention on self-reported levels of mindfulness and psychological distress, as well as the relationship between them. In doing so, the project sought to identify important implications for how to best support service users and identify whether particular facets of mindfulness are associated with more positive outcomes which therefore may warrant more focus within

the group setting. Following discussion with facilitators of the group, this project identified the following research questions:

Primary Aims

1. Is there a reduction in symptoms of distress (as measured by the CORE-OM questionnaire), from the beginning to the end of the Mindful Life group? It is hypothesised that there will be a difference in the self-reported levels of distress at the two timepoints.

2. Is there an increase in mindfulness-based skills (as measured by the Five Facet Mindfulness questionnaire), from the beginning to the end of the group? It is hypothesised that there will be a difference in the self-reported mindfulness-based scores at the two timepoints.

Secondary Aims

1. Is there an association between mindfulness skills and distress? It is hypothesised that there will be a relationship between the two variables.

2. Are there particular facets of mindfulness that are associated with more positive outcomes for service users?

Method

Ethical Approval

Data used in this project had been routinely collected and anonymised by a secondary mental health service in the UK. Analyses were conducted as part of a service-related project, approved by the local quality improvement team in the NHS trust (Appendix F).

Participants

Participants were service users within a UK secondary mental health team who had been referred (*n* = 201), assessed and invited to attend a 12-week mindfulness group (*n* = 111) delivered by staff within the service. The group was delivered in person three times per year, with six to ten participants in each group cohort. Attendees were referred by five services and experienced a range of physical and mental health difficulties.

Measures

Those who attended the group were invited to complete two standardised questionnaires at two separate timepoints: once during the first session of the group and again in the twelfth and final session. The measures used included the Clinical Outcomes in Routine Evaluation – Outcome Measure (CORE-OM) (Evans et al., 2000), completed by 38 participants, and the Five-Facet Mindfulness Questionnaire – Short form (Bohlmeijer et al., 2011), completed by 40 participants.

Clinical Outcomes in Routine Evaluation – Outcome Measure (CORE-OM). The CORE-OM (Evans et al., 2000) is a 34-item self-report measure of psychological distress and wellbeing, with focus over the previous week. Responses are recorded on a five-point frequency scale, from 'not at all' (0) to 'most or all of the time' (4), with some items requiring reverse-scoring. The measure consists of four domains which look at psychological wellbeing (four items), functioning (12 items), symptoms (12 items), and risk (six items). Scores are calculated as the mean average for the domain items (between 0 and 4) and presented as a clinical score by multiplying the mean by ten (Connell et al., 2007), with a minimum score of 0 and maximum score of 40 - higher scores indicating a higher severity of symptoms and higher distress. Research has shown CORE-OM to be a reliable (α = 0.75-0.95) and valid measure of psychological distress, with good sensitivity to change (Evans et al., 2002).

Five-Facet Mindfulness Questionnaire-Short Form (FFMQ-SF). The FFMQ-SF (Bohlmeijer et al., 2011) is a 24-item self-report measure of different elements of mindfulness practice, which was shortened from the original 39-item questionnaire (Baer et al., 2006). Responses are reported on a five-point Likert scale, from 'never or very rarely true' (1) to 'very often or always true' (5), with some items being reverse-scored. The questionnaire comprises five domains of mindfulness: non-reactivity to inner experience (five items), observing (four items), acting with awareness (five items), describing (five items), and non-judging of inner experience (five items). The responses are summed to calculate total score. The scores range from 24 – 120, with higher scores representing higher levels of mindfulness. Research has shown FFMQ-SF to be reliable, valid and with good sensitivity to change in adults with depression and anxiety (Bohlmeijer et al., 2011).

Intervention

The group intervention, adapted from a manualised MBCT group (Segal et al., 2012), consisted of 12-weekly sessions of 1.5 hours each. Each session was routinely facilitated by two experienced members of staff (a clinical psychologist and nurse specialist), often with a non-qualified observer, such as a trainee clinical psychologist or

trainee psychiatrist. The same structure was used for each of the 12 sessions, beginning with a five to 15-minute guided mindfulness practice to welcome everyone to the group. A different practice was introduced each week, in line with the theme of the group. The practice was followed by a check-in and review of the homework, with attendees encouraged to share their experience of, or difficulties with, mindfulness practices over the week. The majority of time in each session focused on a group discussion of the weekly topic (Table 2.1) and those attending were invited to contribute as much or as little as they liked. Each session ended with a final mindfulness practice and handouts were provided to participants as a reminder of session content and homework suggestions.

Table 2.1

Topics Covered Across the Twelve-Week Mindfulness Group

Session	Theme	Suggested Homework Practice
1	What is mindfulness	Eating mindfully
2	Human emotion systems	Noticing doing something good for self/others and deep breathing practice
3	Tricky brain, inattention, and attention	Inattention and attention (grounding and soothing)
4	Mindfulness of body	Non-judgement
5	Acceptance	Acceptance
6	Mindfulness of breath	Breath
7	Mindfulness and pain	Body and pain
8	Mindfulness of action	Action
9	Mindfulness of thoughts	Thoughts
10	Mindfulness of emotions	Emotions
11	Mindfulness in relationships	Relationships
12	Compassion	Compassion/kindness

Analysis Plan

Of the 111 participants invited to attend the group, 70 participants completed the intervention. From this number, 50 participants completed at least one paired outcome measure. The data from participants who completed paired mindfulness ratings (n = 40) was analysed using a non-parametric test (Wilcoxon signed ranks test), comparing the differences in mean score between pre and post intervention timepoints (primary aim one). For paired CORE-OM data (n = 38), a further Wilcoxon signed ranks test was

conducted to analyse differences in mean scores for pre- and post- distress ratings (primary aim two). The non-parametric tests were appropriate due to the degree of skew observed within the distribution of the data. The analysis also included calculations of effect sizes (r).

To test secondary aims and look at the relationship between mindfulness and distress, data was gathered from participants who completed both sets of outcome measures (*n* = 28). Change score calculations were conducted by subtracting pre-intervention scores from the post-intervention scores for mindfulness and distress ratings. Non-parametric correlational analyses (Spearman's Rho) were conducted based on these change scores to investigate the relationship between the overall clinical CORE-OM score and the total FFMQ-SF score (secondary aim one) and the five facet subscales of the FFMQ-SF (secondary aim two). The hypotheses were all two-tailed, with a statistical significance value set to p=0.05.

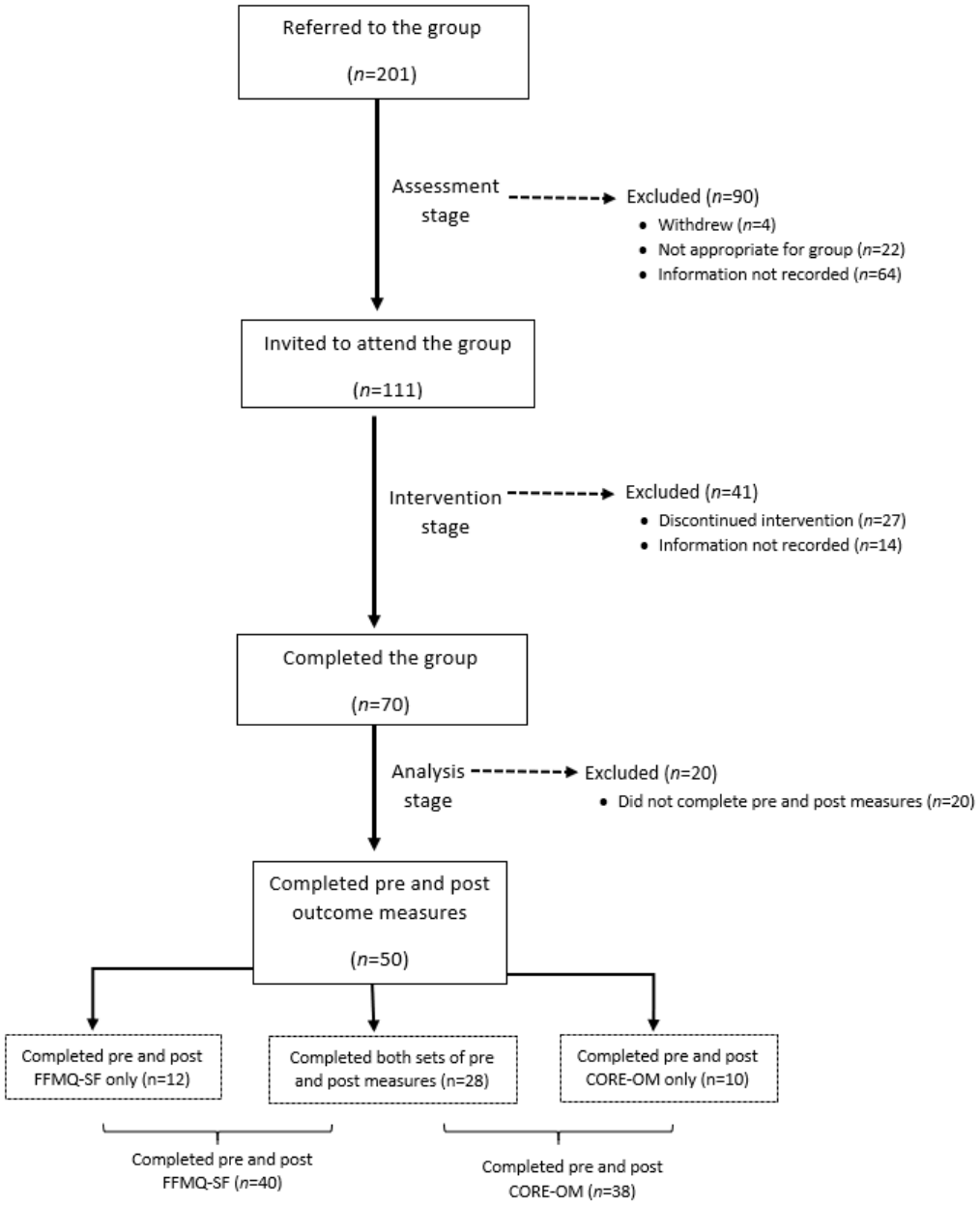
Results

From January 2015 to February 2020, there were a total of 14 cohorts of the Mindful Life group. During this time, 201 people had been referred to the group – each of these referrals were assessed by facilitators to determine if they were appropriate for the group. Those considered appropriate, based on facilitator judgement, were invited to start (*n* = 111) in the next group cohort. Overall, 70 participants went on to complete the group, whilst 27 dropped out, and completion status was not recorded for the remainder of participants (*n* = 14). Information on withdrawal reasons was not recorded for many of those who dropped out (*n* = 16) however those who offered a reason were most likely to drop out due to ill health (*n* = 6). Other reasons for withdrawal included anxiety (*n* = 3) and bereavement (*n* = 2*)*. In terms of mean average, participants attended 7.95 sessions (range 0-12), with 10 sessions being the mode average.

Of those who completed the group (*n* = 70), 71.4% (*n* = 50) completed at least one outcome measure at both the start and end of the intervention and have been included in the analysis (Figure 2.1). At least two people completed paired outcome measures across 13 cohorts of the group (a range of 2 to 7 attendees completed outcome measures in each of these 13 group cohorts).

Figure 2.1

Flow Diagram of Participants, from the Referral Stage to Completion of Group and Outcome Measures



Note. FFMQ-SF, Five Facet Mindfulness Questionnaire – Short Form; CORE-OM, Clinical Outcomes in Routine Evaluation – Outcome Measure

Demographics

The demographics of referrals and those who participated in the group are shown in Table 2.2. The majority of participants were referred by the recovery team (33.8%) although this reduced throughout the assessment stage suggesting that these referrals were more likely to be deemed inappropriate or withdraw. By the start of the group, those held within the later life team made up majority of attendees (33.3%) and were more likely to complete outcome measures at beginning and end of the group intervention (36%). Participants within the EI team and primary care liaison service made up the minority of the referrals (5.5% and 0.5%, respectively) and were least likely to complete both pre and post outcome measures (8% and 0%, respectively) – although it should be noted that the referral from the primary care team appeared to be an anomaly as referrals were not usually accepted from this service.

Table 2.2

Demographics of Participants at Each Stage of the Group Intervention

Participant characteristic	Referrals to the group *n = 201*		Those invited to attend group *n = 111*		Attendees with outcome measures *n = 50*	
Age						
Mean	50.27 years		51.39 years		52.12 years	
Standard Deviation	16.22		16.38		16.63	
Range	21 – 80		21 - 80		21 - 80	
	n	%	*n*	%	*n*	%
Age category						
18 – 39	46	22.9	27	24.3	15	30
40 – 64	55	27.4	31	27.9	13	26
65+	50	24.9	40	36	21	42
Unknown	*50*	24.9	13	11.7	1	2
Gender						
Male	71	35.3	44	39.6	25	50
Female	12	59.7	67	60.4	25	50
Unknown	0	5	0	0	0	0
	10					
Referrer						
Psychological Therapies	52	25.9	28	25.2	13	26
Recovery team	68	33.8	27	24.3	12	24
Early Intervention	11	5.5	6	5.4	4	8
Later Life	47	23.4	37	33.3	18	36
Primary Care Liaison Service	1	0.5	1	0.9	0	0
Unknown	22	10.9	12	10.8	3	6
Diagnosis						
Mood disorder	28	12.8	20	16.1	9	14.8
Obsessive-compulsive and related disorders	8	3.7	4	3.2	3	4.9
Anxiety disorder	18	8.3	15	12.1	11	18
Personality disorder	14	6.4	7	5.6	2	3.3
Bipolar and related disorders	22	10.1	9	7.3	2	3.3
Schizophrenia spectrum & other psychotic disorders	18	8.3	11	8.9	8	13.1
Adjustment disorder	1	0.5	1	0.8	1	1.6

Neurocognitive disorder	2	0.9	2	1.6	2	3.3
Other e.g., abuse, anger, awaiting diagnosis	3	1.4	1	0.8	1	1.6
Substance use disorder	1	0.5	1	0.8	0	0
Trauma and stress related disorder	1	0.5	1	0.8	1	1.6
Mixed anxiety / depression	8	3.7	4	3.2	2	3.3
Unknown	94	43.1	48	38.7	19	31.1

The demographics show that people with a wide range of difficulties were referred to the group. As some participants experience comorbid diagnoses, there are more diagnoses (*n* = 218) than participants (*n* = 201). Those with mood disorders, such as depression, were referred to the group most often, however did not consistently complete pre and post outcome measures.

Psychological distress ratings (as measured by CORE-OM)

Whilst average scores showed a slight decrease in levels of overall distress ratings (Table 2.3) from the start (*Mdn* = 17.25) to end of the intervention (*Mdn* = 14.55), with a small effect size, a Wilcoxon signed ranks test showed that these differences were not statistically significant (p>0.05).

Mindfulness ratings (as measured by FFMQ-SF)

Overall mindfulness scores increased from start (*Mdn* = 60.50) to end of the group (*Mdn* = 70.00). Wilcoxon signed rank tests show a statistically significant difference (p<0.01). Similar results were also shown in three of the five facets of mindfulness, including non-reacting, observing and non-judging (p<0.05) – each of these showed small to medium effect sizes.

The final two facets of mindfulness, awareness and describing, did not demonstrate significant differences between start (*Mdn* = 14:00; 14.50, respectively) and end of the group intervention (*Mdn* = 14.00; 15.00, respectively).

Table 2.3

Paired Comparisons of Mindfulness and Psychological Distress Scores

Measure	Pre-intervention		Post-intervention		t	z	p	Effect size (r)
	Mdn	*IQR*	*Mdn*	*IQR*				
CORE-OM (n=38)	17.25	10.23	14.55	9.42	280	1.31	.189	-0.15
FFMQ-SF(n=40)								
Non-reacting	10.50	5.00	13.00	6.00	438	2.82	.005*	0.35
Observing	12.50	5.00	13.50	4.00	413	2.39	.017*	0.29

Awareness	14.00	6.00	14.00	5.00	355.5	1.00	.316	0.12
Describing	14.50	7.00	15.00	7.00	284.5	0.39	.699	0.05
Non-judging	11.00	5.00	14.00	7.00	561.5	3.18	.001**	0.37
Overall	60.50	19.00	70.00	20.00	649	3.22	.001**	0.36

Note. Mdn, Median; IQR, Interquartile Range; CORE-OM, Clinical Outcomes Routine Evaluation-Outcome Measure; FFMQ-SF, Five Facet Mindfulness Questionnaire-Short Form.

*p < .05

**p < .01

Correlations

Whilst a significant correlation was not observed between change scores of overall mindfulness and overall distress, a trend for significance was noted (p = .059). The change score for the non-reactivity to inner experience facet of FFMQ-SF, however, showed a significant negative correlation with the total change score captured in the CORE-OM (p <0.01), suggesting that as non-reactivity-based mindfulness skills increase, distress decreases. No significant relationships were found between change scores for the other four facets of the mindfulness questionnaire (observing, acting aware, describing, non-judging) and overall changes in distress (p > .05). A summary of correlation coefficients between the CORE-OM clinical score and FFMQ-SF overall and subscales are shown in Table 2.4.

Table 2.4

Correlations Between Change Scores of Mindfulness and Psychological Distress (n=28)

	FFMQ-SF					
Variable	Non-reacting	Observing	Awareness	Describing	Non-judging	Overall
CORE-OM						
Correlation	-.68**	.038	.058	-.03	-.32	-.36
Significance	.000	.848	.770	.881	.101	.059

Note. CORE-OM, Clinical Outcomes Routine Evaluation-Outcome Measure; FFMQ-SF, Five Facet Mindfulness Questionnaire-Short Form.

*p < .05

**p < .01

Discussion

The aim of this study was to evaluate the short-term effect of a mixed-age and -presentation mindfulness group intervention on self-reported psychological distress and mindfulness-based skills. The findings show that mindfulness-based skills increased from start to end of group, with small to moderate effect. Significant improvements were noted in overall levels of mindfulness as well as three of the five facets of mindfulness, which included non-reacting, observing and non-judging of inner experiences. For the acting aware facet, no change was observed over the course of the group, whilst the domain of describing showed a slight but non-significant increase. Similarly, a decrease in the average ratings for psychological distress was demonstrated, indicating small reductions in distress, although this did not prove to be a statistically significant change.

Based on previous research, it was expected that both distress and mindfulness would show significant differences between the two timepoints, as has been shown in mindfulness-based interventions for both clinical (Baer et al., 2012) and non-clinical populations (Virgili, 2015). Whilst not-significant, results from this study are broadly consistent with previous research in the use of mindfulness-based interventions for heterogenous outpatient groups, for example Ree and Craigie (2007) found statistically significant improvements in symptoms of stress and mindfulness, with a small effect size. The results in this paper do however contrast with a recent evaluation of a clinical-mindfulness group within Sussex Partnership NHS Trust, which found significant improvements in symptoms of anxiety and depression and non-significant increases in mindfulness skills (Strauss & Hughes, 2019). A key difference may be the use of symptom-specific measures in the Sussex evaluation which would not have been appropriate given the heterogeneity of mental health presentations included in the Mindful Life group.

Given that ratings of overall mindfulness increased over the course of the group, it indicates that participants engage with some aspects of mindfulness, at least within the weekly group setting. Previous studies have demonstrated a relationship between home-practices of mindfulness and clinical outcomes (Crane et al., 2014; Parsons et al., 2017) and whilst this was encouraged throughout the intervention, the service did not collect information on engagement in at-home practice. Armstrong's (2019) qualitative review of the subjective experiences of 8 attendees of the Mindful Life group noted that participants varied in their levels of commitment and motivation to engage in home-practice, with some reflecting that continued practice may have supported further changes and most participants reporting in-session practice to be easier than home-practices. It would be

interesting for the service to investigate the engagement in home practices in future groups as it is possible this goes someway to explaining the small but non-significant changes in distress levels.

Further research investigating subjective experiences of mindfulness-based interventions noted that some participants described acceptance of thoughts and emotions as uncomfortable and became more aware of the impact of difficulties on their lives (Wyatt et al., 2014). This is supported by Armstrong's (2019) qualitative evaluation of the Mindful Life group whereby some participants found practices frightening or uncomfortable and worried about the loss of usual coping strategies. The Mindful Life group encourages participants to turn towards distress rather than engaging in experiential avoidance which is often linked with mental health difficulties (Sagui-Henson, 2017) and may therefore be an ingrained coping strategy for those in secondary mental health services. As mindfulness-based interventions seek to reduce cognitive avoidance (Eisendrath et al., 2016), this may result in participants becoming more aware of internally distressing experiences (Mahoney et al., 2015) - it is possible that some attendees may have found this difficult, resulting in non-significant changes in overall distress. Additionally, stressful life events and personal circumstances were not recorded over the duration of the group and so it is not possible to investigate whether this influenced self-reported levels of distress.

The secondary aims of the study were to investigate the relationship between distress and mindfulness skills overall, alongside the association between distress and the five facets of mindfulness measured within the questionnaire. Whilst a small but non-significant relationship was shown between increased overall levels of mindfulness and reduced distress, the domain of non-reactivity to inner experience showed a significant inverse relationship with distress. Whilst causation cannot be determined, this suggests that as elements of non-reactivity increase, distress reduces and is in line with previous research of the mechanisms behind mindfulness interventions (Gu et al., 2015).

Similar to work by Garland and colleagues (2013), through showing the associations between distress and each of the FFMQ-SF facets, alongside the overall FFMQ-SF score, the study demonstrates an implication for understanding how the different elements of mindfulness relate to a reduction in psychological distress. The analyses in this study suggest that non-reactivity to inner experience, the ability to notice thoughts without trying to change or react to them, is a key component to distress-reduction. Similar implications have been suggested in previous research with non-reactivity showing an important inverse relationship with symptoms of post-traumatic

stress disorder (Kalill et al., 2014), depression (Barnes & Lynn, 2010) and non-suicidal self-injury (Caltabiano & Martin, 2017).

Additionally, in research which mapped facets of mindfulness with depression and anxiety (Desrosiers et al., 2013), a significant inverse relationship was found between non-reactivity and general distress in anxiety and depression symptoms, whilst no association was observed with acting aware. In contrast, recent research by Chien and colleagues (2020) found that “observing” and “acting with awareness” were key to supporting positive outcomes for people with psychosis. This raises a question of whether there are diagnosis-specific relationships between different elements of mindfulness, as suggested by Didonna and colleagues (2019), which was not captured within this research. It may be helpful for future research to systematically investigate this which could be a key consideration in delivering mixed-presentation mindfulness groups in terms of emphasising different practices depending on participant difficulties.

Strengths and Limitations

Whilst self-reported ratings of mindfulness-based skills appeared to increase from the start to end of the group, the lack of control group within this project limits the conclusions that can be drawn. It is possible that increased ratings are explained by social desirability effects (Banerjee et al., 2018) and the relationships established with group facilitators. Previous research (Allen et al., 2009) has also indicated that group processes may interact with skills in mindfulness-based interventions which further complicates the findings and may be reflected in outcome measures.

A strength of this study is that demographics and outcome measures were collected by the service for five years and from several cohorts of the mindfulness intervention, providing data from a large number of participants. This provides a clear overview of the demographics of the referrals to the group; however, it should be noted that there are gaps in the data as not all information was recorded for each participant, with over 30% of diagnosis data missing from those who completed the group. In addition, certain demographics such as ethnicity have not been collected and thus it is difficult to comment on the degree to which the sample reflects the local population, and therefore how generalisable the findings are.

Furthermore, the study looked at only the participants who had completed at least one paired outcome measure. Encouragingly, this was over 70% of those who completed the group, with demographics broadly reflecting those invited to attend the group. However, there were fewer participants who completed both sets of outcome measures at

the two timepoints; therefore, the correlational analyses are based on a subsample of participants. It is possible that those who were more engaged with the group were more likely to complete both outcome measures at the end of the intervention. It is recommended that all attendees of the group are supported to complete the self-reported questionnaires as this may help to strengthen the service research. It would also be beneficial for the service to consistently collect information related to those who withdraw from the intervention, as this may provide a greater understanding of the barriers. Finally, the study has not investigated the impact of home-practices and it is unclear whether this could contribute to more statistically significant changes in distress levels – in the future, it may be helpful for the service to collect information on the frequency and type of home practice that participants engage in as this may create further reductions in participant distress.

Recommendations and Implications for the Service

The findings and recommendations have been discussed with the facilitators of the Mindful Life group via a virtual meeting and demonstrated in a summary report. The facilitators will also share these findings with the management team of the service.

The demographics demonstrate that the EI in psychosis team made the fewest referrals to the mindful life group over the five years (5.5%). It is possible that this may be due to unsubstantiated concerns regarding the relationship between mindfulness and risk of increased symptoms in psychosis (Jacobsen et al., 2019); however, this is not consistent with recent research which indicates that mindfulness-based interventions can be beneficial for this population (Aust & Bradshaw, 2017; Jacobsen et al., 2019). Following discussion with group facilitators, it was noted that clinicians in the EI service may be drawn to other therapeutic approaches which may impact on the number of referrals made to the group. It may be useful for information regarding the mindfulness group and outcomes to be shared with the EI service to support future referrals, or to consider whether additional collaboration or adaptations are required to support those with first-episode psychosis.

It would be interesting for a future service-related project to track the participants following the completion of the Mindful Life group; this may be beneficial to look at the number of participants that are discharged from services following the intervention as well as those that go on to access further therapy within the services and which therapies attendees are most likely to access following the group. This analysis may usefully support understanding of the outcomes of the intervention which may not be captured

through self-rated distress and may be better captured through the impact on service demand and waiting lists for other therapeutic activities.

The evaluation suggests that non-reactivity to inner experiences is a key component of mindfulness which correlates with reduced distress. As such, it is recommended that further mindfulness practices are introduced to support participants to develop a non-reactive stance to their inner experiences. This may support further reductions in distress, evidenced through the continued use of outcome measures. Recognising non-reactivity as a key facet of mindfulness may also support participants to develop more specific therapeutic goals for the intervention.

Less than half of those who completed the group completed both sets of outcome measures at the two timepoints, however participants were more likely to complete one outcome measure at the beginning and end of the group. The facilitators may wish to explore ways to support participants to fill out both sets of outcome measures in order to continue evaluating the impact of the intervention. Alternatively, it may be that the two questionnaires together are too high a burden for attendees and shorter questionnaires may be more appropriate. From discussion with the facilitators, it was queried whether someone could take on the role of completing measures on the phone following completion of the group. Previous research has also recommended that withdrawal reasons are recorded to understand the difficulties and barriers to attending mindfulness-based interventions (Nam & Toneatto, 2016) – this information may be helpful for the service to support solution-focused discussions in assessment.

In line with previous qualitative research with participants of the Mindful Life group (Armstrong, 2019), it is recommended that information is gathered on the use of home-practice as this may support improvements in distress. Future research with the service could explore frequency and impact of home-practices, as well as which practices are preferred. This may further support discussions regarding commitment and engagement within the assessment stage and the development of goals for service users.

Finally, given the current context of the COVID-19 pandemic there is a move towards remote delivery of interventions. Previous research has shown that online mindfulness interventions can be effective in reducing psychological distress in clinical populations (Compen et al., 2018). The facilitators of the group note that the intervention has been transferred to a remote environment and it may be useful for a future service-related project to compare outcomes from in-person and remote delivery.

In conclusion, the study offers tentative evidence that a 12-week mindfulness group intervention, delivered within a secondary mental health service, increases mindfulness-based skills and slight reductions in psychological distress. Not reacting to inner experiences is highlighted as a key mindfulness skill, linked with reductions in psychological distress. Further improvements in self-reported distress may therefore be supported through increased focus on practices which help clients to develop a non-reactive approach, and these should be emphasised throughout the intervention.

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Main Research Project

A mixed-methods study of fatigue in young people with neuromuscular conditions:

"It limits how much I can do, it limits the quality of how much I can do, and it's a reminder of your disability which [affects] mental health"

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Proposed Journal: Journal of Neuromuscular Disorders (Appendix G)
The Journal of Neuromuscular Disorders publishes research related to all neuromuscular conditions in children and adults, with a word limit of 2500 – 6000 words.

A mixed-methods study of fatigue in young people with neuromuscular conditions: "It limits how much I can do, it limits the quality of how much I can do, and it's a reminder of your disability which [affects] mental health"

Abstract

Neuromuscular conditions are progressive disorders associated with muscular weakness. Fatigue is a common symptom in adults with specific types of neuromuscular conditions, however, less is known about fatigue in young people with neuromuscular conditions. This study aimed to explore prevalence and severity of fatigue, and associated factors, alongside lived experiences of fatigue in young people with neuromuscular conditions. A cross-sectional mixed methods design was used; thirty-three participants with ten neuromuscular conditions completed an online survey including measures of fatigue, mood, sleep quality, responses to symptoms, social functioning, physical functioning, and quality of life. Data were analysed with bivariate correlations and stepwise linear regression. Twelve participants completed follow-up interviews to explore fatigue experiences, analysed with Reflexive Thematic Analysis. In total, 24% of participants reported significant fatigue, which was associated with sleep quality ($r = -.39$, $p = .025$) and depression symptoms ($r = .40$, $p = .022$). Five themes were constructed from interviews capturing the far-reaching impact of fatigue and difficulties in managing it. Quantitative measures and qualitative interviews indicate that fatigue is a common experience for young people with neuromuscular conditions and is strongly associated with feelings of depression. Implications for services, alongside limitations of this study and recommendations for future research are discussed.

Introduction

The term ‘neuromuscular disorder’ (NMD) refers to a heterogenous group of rare conditions characterised by impairments in the functioning of muscles and nerve structure. Many of these conditions are genetic and progressive, causing a decline in muscle strength and leading to difficulties with mobility and complications with cardiovascular and respiratory functioning (Rodino-Klapac et al., 2013; Royal College of Nursing, 2019). Whilst age of onset varies (Dowling et al., 2018), previous research indicates that the overall population prevalence for most neuromuscular conditions is between 1 and 10 per 100,000 (Deenen et al., 2015), however recent prevalence ratings (Carey et al., 2021) suggest the number of people in the UK living with muscle-wasting conditions may be higher than previously thought, indicating increased life expectancy due to advances in medical care (Rodger et al., 2015).

Despite medical advances, there is currently no cure for neuromuscular conditions and the rate of progression varies. As such, continued support from multidisciplinary teams is required (Bushby et al., 2010). An all-party parliamentary review for muscular dystrophy (2018) indicated the need for improved access to psychological support, however there is currently limited research regarding efficacy of psychological interventions for this population (Walklet et al., 2016) and further research is required (Graham et al., 2015).

Fatigue

Whilst fatigue is a subjective experience (Loades & Chalder, 2020), it is often described as an overwhelming feeling of exhaustion and lack of energy which affects day-to-day activities (de Vries et al., 2010). In chronic fatigue research, adolescents have described feeling ‘heavy’ and ‘drained’ (Parslow et al., 2018, p. 2). Similar descriptions have been reported by adolescents with cancer who described feeling physically and mentally drained (Hockenberry-Eaton & Hinds, 2000). In the same study, oncology staff recognised fatigue through changes to motivation, affecting participation in health and social activities. Research with adolescents with chronic fatigue syndrome has demonstrated the far-reaching impact of persistent fatigue, including difficulties with social functioning (Ali et al., 2019), attendance at school (Knight et al., 2018), and learning and memory (Sulheim et al., 2015).

To date, there has been limited research regarding fatigue within young people with NMD (El-Aloul et al., 2020). Fatigue, has however, been recognised as a difficulty for young people with other chronic health conditions (Nap-Van Der Vlist et al., 2019) and is

considered a key symptom in adults with NMD (Wokke, 2007), with over 60% reporting it as their most disabling symptom (Kalkman et al., 2005). For adults with NMD, fatigue contributes to difficulties with social functioning (Kalkman et al., 2005), occupational functioning (Lindsay et al., 2019), and quality of life (Pangalila et al., 2015).

Previous research (Kalkman et al., 2007) proposed a model to explain experiences of fatigue for adults with NMD which highlights that physical activity, muscle strength and pain contribute to fatigue and disrupt day-to-day functioning. This model, however, does not consider psychological features which may also contribute to fatigue. In contrast, the cognitive behavioural model of fatigue (Surawy et al., 1995) recognises the role of psychological factors, such as anxiety, depression, thoughts and behaviours (Loades & Chalder, 2020), as well as biological and environmental aspects, and has been developed within a paediatric population with chronic illness. This model identifies a vicious cycle in which oscillations between surges of activity and ineffectual rest ('boom and bust' cycles) perpetuate difficulties with fatigue.

Whilst fatigue is documented in the adult NMD literature, the progressive nature of the conditions means that this cannot be generalised to young people without more research. Recent studies have started to explore the difficulties that young people with neuromuscular conditions have with fatigue, however this research has largely focused on specific conditions, such as Charcot-Marie-Tooth disease (Jagersma et al., 2013) and Duchenne muscular dystrophy (DMD) (El-Aloul et al., 2020; Wei et al., 2017), and has shown that fatigue is associated with reduced quality of life (Bhullar et al., 2018; Jagersma et al., 2013; Wei et al., 2016). El-Aloul and colleagues (2020) demonstrated that fatigue severity in young males with DMD was similar to levels reported by paediatric cancer populations. However, as DMD only affects males it is not yet understood as to whether this is a similar experience across the paediatric NMD population. The study recommended further research, including qualitative studies to explore severity and impact of fatigue for this population.

Aims and Hypotheses

This study aims to advance the evidence-based by investigating the following research questions:

1. What is the prevalence of fatigue within young people with neuromuscular conditions? Based on previous research which demonstrates that fatigue is prevalent amongst those with chronic health conditions (Nap-Van Der Vlist et al., 2019), it is hypothesised that fatigue will be a common experience within this population.

2. What factors are associated with fatigue in young people with neuromuscular conditions? Previous studies with those with specific neuromuscular conditions (El-Aloul et al., 2020; Bhullar et al., 2018) show that fatigue is linked to symptoms of depression, sleep, and quality of life. It is hypothesised that these factors will show the strongest relationships with fatigue in this study.

3. What are young people's experiences of fatigue within neuromuscular conditions and how does it impact on their life? To date, there has been limited opportunities for young people with NMD to have their voice heard within research, as such this study also aims to qualitatively explore young people's experiences of fatigue.

Method

Design

A cross-sectional mixed method design was used.

Participants

Eligible participants were recruited through third-sector organisations, social media, and mailing lists. Participants were included if they were aged 10 - 24, with a self-reported neuromuscular diagnosis, able to understand and complete measures in English, and lived in the UK. Participants were not eligible to take part if they were unable to complete questionnaires, either individually or with support from a parent, carer, or researcher.

Measures

Socio-demographic information was collected at the start of the survey; this included age, gender, ethnicity, type of neuromuscular condition, co-morbidities, medication, ambulatory status, and wheelchair use. Standardised self-report measures were used to assess fatigue, symptoms of depression and anxiety, physical functioning, health-related quality of life (HRQOL), sleep, social functioning, and cognitive and behavioural responses to symptoms.

Fatigue was measured using the Chalder Fatigue Questionnaire (CFQ) (Chalder et al., 1993); an 11-item self-report measure of fatigue, evidenced for those with chronic fatigue syndrome as well as those with chronic health conditions (Cella & Chalder, 2010). Responses are rated on a four-point scale, with each item scored 0-3 and overall scores ranging from 0 – 33. Higher scores indicate higher levels of fatigue. Previous research with adolescents with HIV (Coetzee et al., 2018) and chronic fatigue syndrome (Lloyd et

al., 2012) has established a threshold of ≥ 18 to identify those for whom fatigue is a significant problem. The measure has been shown to be a valid and reliable measure of fatigue in young people (Bould et al., 2013; Collin et al., 2015) and in adults with neuromuscular conditions (Hoffmann et al., 2016; Laberge et al., 2005). Cronbach's alpha was .74, indicating acceptable internal consistency.

The short version of the Revised Children's Anxiety and Depression Scale (RCADS-25) (Ebesutani et al., 2012) was used to measure symptoms of anxiety (15 items) and depression (10 items). Responses are rated on a four-point scale ranging from 0 (Never) to 3 (Always) and higher scores suggest increased symptoms. Psychometric properties are lower than that of the original full-length version (Chorpita et al., 2005), however acceptable validity and reliability have still been shown with young people (Ebesutani et al., 2012; Piqueras et al., 2017). Cronbach's alpha was .93, indicating good internal consistency.

Physical health was measured using the physical functioning scale of the Short-Form 36 (SF-36 PFS) which consists of 10-items related to how limited participants feel in day-to-day physical tasks (Ware & Sherbourne, 1992). Responses are rated using a 3-point scale and scored as 0 (limited a lot), 5 (limited a little), or 10 (not limited at all); higher scores demonstrate better physical functioning. The full-length SF-36 is a valid and reliable measure in adults with neuromuscular conditions (Boyer et al., 2006) and the physical subscale has been shown to be reliable and valid with adolescents with chronic fatigue (Loades, Vitoratou, Rimes, & Chalder, 2020). Cronbach's alpha was .95, indicating good internal consistency.

The Cognitive Behavioural Responses Questionnaire – Short version (CBRQ-S) was used to measure cognitive and behavioural responses to symptoms (Ryan et al., 2018). Eighteen items were rated on a five-point scale, from 0 (never or strongly disagree) to 4 (all the time or strongly agree), with higher scores indicating more unhelpful responses. Good validity and reliability has been demonstrated in adolescents with chronic fatigue (Loades, Vitoratou, Rimes, Ali, et al., 2020). In this sample, Cronbach's alpha was .86, indicating good internal consistency.

The School and Social Adjustment Scale (SSAS) (Loades et al 2020a), adapted from the adult-based Work and Social Adjustment Scale (Mundt et al., 2002) for use with adolescents, was used to measure social functioning. Participants are asked to rate 5 items on an eight-point scale related to how much they believe their illness affects activities. Scores range from 0 – 40 and higher scores suggest more difficulties. The SSAS is a reliable and valid measure for young people aged 11 to 18 with chronic fatigue

(Loades, Vitoratou, Rimes, & Chalder, 2020). Cronbach's alpha was .84, indicating good internal consistency.

Health-related quality of life was measured using Kidscreen-27 (Ravens-Sieberer et al., 2005) which consists of 27 items across five domains of physical wellbeing, psychology wellbeing, autonomy, social support and school environment. Responses are rated on a five-point scale, with some items reverse scored. For each domain, T-scores are calculated which have a mean of 50 and standard deviation of 10 (Ravens-Sieberer, 2006). A total score is calculated by collating all responses and higher scores indicate better HRQOL. This has been shown to be a valid measure of HRQOL in children and adolescents (Ravens-Sieberer et al., 2007); Cronbach's alpha in this sample was .87, indicating good internal consistency.

The Adolescent Sleep-Wake Scale-Short Version (ASWS-S) was used to measure sleep quality (Essner et al., 2015) and consists of 10 items, adapted from the original 28-item measure (LeBourgeois et al., 2005). Responses are rated on a six-point scale, from 1 (always) to 6 (never) of how often sleep difficulties have occurred over the last month. A total score is then calculated based on the mean of all items, with scores ranging from 1-6 and higher scores reflect better quality sleep. Research shows this to be a reliable and valid measure for those with and without health conditions (Essner et al., 2015; Sufrinko et al., 2015), and in young adults as well as adolescents (Huber et al., 2020). Cronbach's alpha in this sample was .75, which indicates acceptable internal consistency.

All measures were self-report, although parents and caregivers were invited to help young people complete the survey.

A semi-structured interview guide (Appendix H) was created based on previously published literature (Carroll et al., 2016; Czuber-Dochan et al., 2013), and consisted of open-ended questions on the experience, meaning and impact of fatigue.

Procedure

Those who saw the study advert and were interested in taking part were invited to follow an online link to the Qualtrics platform where age-appropriate information about the project and consent forms were presented. For those aged under 16 years, parents or carers were asked to provide informed consent along with assent sought from the child. Participants aged 16 and above were invited to provide their own consent. Additionally, participants were invited to consent to contact for a follow-up interview. Those who participated were entered into a prize draw to win one of three vouchers.

Through Qualtrics, participants were asked to complete a battery of standardised questionnaires, alongside demographic information. Participants completed the questionnaires between February 2021 and February 2022. Follow-up interviews were completed within the same time frame, using Microsoft Teams. The interviews were designed to take approximately 30 minutes to reduce research burden for participants and avoid exacerbating difficulties with fatigue.

To ensure the information and measures used within this study were age-appropriate, two young people with lived experience of fatigue piloted the online questionnaires and gave input into the design of participant information sheets, consent forms and debrief forms. As part of this pilot phase, it was decided that questionnaires would be presented in a consistent order so as to avoid ending on items that may be upsetting for participants, such as mood-based questions.

Ethical approval (Appendix I) for the study was obtained from the Psychology Research Ethics Committee at the University of Bath (Reference Number: 20-240).

Analysis Plan

Analysis followed an integrated mixed methods approach to answer three research questions:

1. What is the prevalence of fatigue within young people with neuromuscular conditions? Measured with quantitative data based on the CFQ.

2. What factors are associated with fatigue in neuromuscular conditions? Based on tests for normality, mean (*M*) and standard deviation (*SD*) or median (*Mdn*) and interquartile range (*IQR*) were calculated to describe data. A power analysis calculated using G*Power (Faul et al., 2009), required a sample size of 84 participants to detect a medium effect size (r=.3) for a correlation coefficient with 80% power, an alpha of 0.05, and for a two-tailed test. Given that this sample was not statistically powered, exploratory analyses were conducted. Bivariate pairwise correlations, using Spearman's rank correlation coefficient, between fatigue and variables of interest (age, depression, anxiety, sleep, physical functioning, social functioning, HRQOL, cognitive and behaviour responses) were calculated and strength of association noted (Ratner, 2009). An exploratory stepwise multiple linear regression was conducted to explore which variables of interest (independent variables) explain the variance in fatigue (dependent variable).

These were conducted using pairwise exclusion. All statistical analyses were conducted with SPSS and a two-sided significance level of p=0.05.

3. What are young people's experiences of fatigue within neuromuscular conditions and how does it impact on their life? Based on qualitative data, drawing on quantitative and demographic information, and analysed using a reflexive thematic analysis (RTA) (Braun & Clarke, 2006, 2019, 2020).

A critical realist ontology framework was adopted which assumes that reality is mediated by one's resources and socially located knowledge (McEvoy & Richards, 2006). A contextual approach to epistemology was adopted alongside this which highlights the role of context and the position of the researcher in the development and interpretation of knowledge. This approach appeared most compatible with the combined quantitative and qualitative methods used for understanding experiences of fatigue (Maxwell & Mittapalli, 2010). A primarily deductive approach was taken with influence from previous research on experiences of fatigue for those with other chronic health conditions (Carroll et al., 2016; Parslow et al., 2018). Additionally, a degree of inductive analysis was included to ensure that participant voices and meanings were held within themes. Similarly, predominantly semantic codes were constructed based on meaningful information communicated by participants, however latent codes were also included when there was meaningful interpretation by the researcher.

As is required in RTA, we acknowledge that the position of the researcher influences knowledge and interpretation and the 'social GRACES' framework was considered (Burnham, 2012; Roper-Hall, 1998). The lead researcher identified as a white British female and was training to become a clinical psychologist. The researcher did not identify as having a chronic health condition but had periods of experiencing unexplained and post-viral fatigue. This, alongside previous experiences of working within paediatric hospitals, could leave the researcher vulnerable to making assumptions about the impact of fatigue for this group of young people. The use of quantitative data, reflexive discussions, and a second coder for qualitative information was included to challenge these assumptions. The second coder did not identify as having a chronic health condition and was not familiar with fatigue. The primary supervisor was a clinical psychologist with experience of working with adolescents with fatigue in both physical and mental health contexts and had personal experience of post-viral fatigue.

The RTA analysis followed a series of six stages as outlined in previous research (Braun & Clarke, 2006; Campbell et al., 2021), including familiarisation with data, coding,

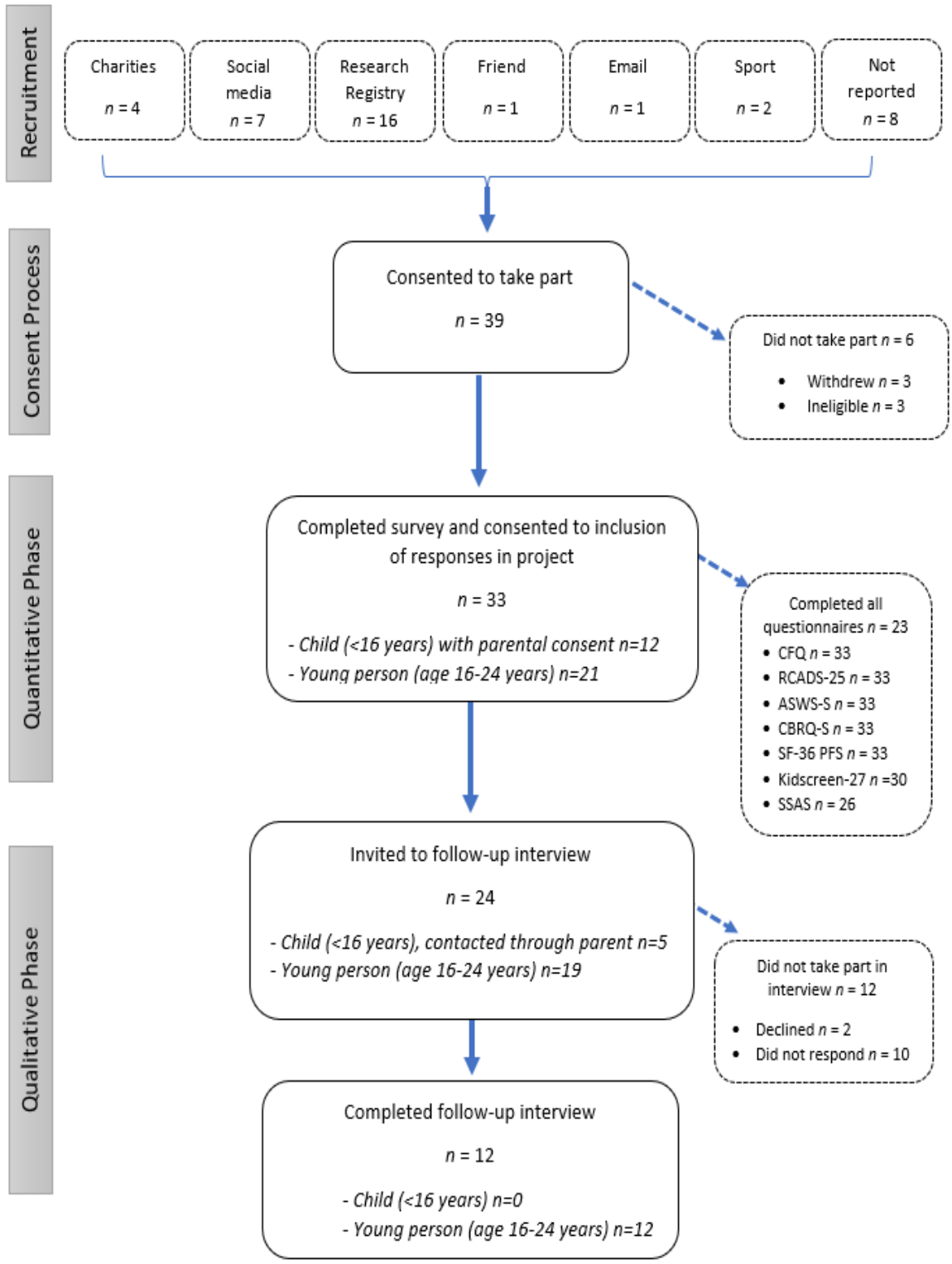
generating themes, reviewing themes, defining themes, and write up. As guided by the RTA process, reflexive conversations were held between the researcher, second coder and primary supervisor. A reflective journal was also used following interviews and throughout the coding process.

Results

Thirty-nine participants consented to take part; three did not meet inclusion criteria and were screened out, whilst a further three participants withdrew without responding to any questions (Figure 3.1).

Figure 3.1

Flow Diagram of Participants at Each Stage of the Project



Note. CFQ, Chalder Fatigue Questionnaire; RCADS-25, Revised Children's Anxiety and Depression Scale; ASWS-S, Adolescent Sleep-Wake Scale-Short Version; CBRQ-S, Cognitive Behavioural Response Questionnaire – short version; SF36 PFS, Short-Form 36 Physical Functioning Subscale; SSAS, School and Social Adjustment Scale

Thirty-three participants completed questionnaires. Mean age was 18.33 years (*SD*=4.2; range 12-24). Participants were mostly British (81.8%) with ten different neuromuscular conditions and myotonic dystrophy was most common (27.3%). Of the 24 participants who consented to follow-up interview, 12 were contactable and completed interviews. Mean age was 21.83 years (*SD*=2.48, range 16-24), and the most common NMD was facioscapulohumeral muscular dystrophy (25%). Demographics and clinical characteristics are shown in Table 3.1.

Table 3.1

Demographic and Clinical Characteristics of Participants, Including Those who Completed the Survey (Quantitative Analysis) and the Subsample who Completed Interviews (Qualitative Analysis)

Participant characteristic	Participants who completed survey *n = 33*		Subsample of participants who completed interview *n = 12*	
Age				
Mean (SD)	18.33 (4.20)		21.83 (2.48)	
Range	12 – 24		16 – 24	
	n	%	*n*	%
Gender				
Male	15	45.5	3	25
Female	18	54.5	9	75
Ethnicity				
White British / English / Welsh / Scottish / Northern Irish	27	81.8	8	66.7
White and Black Caribbean	1	3.0	0	0
White and Black African	2	6.1	1	8.3
African	1	3.0	1	8.3
Pakistani	1	3.0	1	8.3
White and Asian	1	3.0	1	8.3
Diagnosis				
Limb-girdle muscular dystrophy	4	12.1	0	0
Duchenne muscular dystrophy	3	9.1	1	8.3
Myotonic dystrophy	9	27.3	2	16.7
Spinal muscular atrophy	5	15.2	2	16.7
Congenital myopathy	2	6.1	2	16.7
Charcot-Marie Tooth disease	1	3.0	1	8.3
Congenital myotonic dystrophy	2	6.1	0	0

Becker muscular dystrophy	1	3.0	1	8.3
Congenital muscular dystrophy	1	3.0	0	0
Facioscapulohumeral muscular dystrophy	5	15.2	3	25.0
Wheelchair usage				
Not a wheelchair user	21	63.6	7	58.3
Wheelchair user	12	36.4	5	41.7
- *0-25% over the last week*	*1*		*0*	
- *25 – 50%*	*1*		*1*	
- *50 – 75%*	*3*		*0*	
- *75 – 100%*	*7*		*4*	
Other medical conditions/diagnoses				
No	28	84.8	11	91.7
Yes	5	15.2	1	8.3
Medication				
No	16	51.5	6	50
Yes	17	48.5	6	50

Quantitative Analysis (n=33)

Fatigue Severity (CFQ). Using Likert-scoring (0-3), fatigue severity ranged from 2 – 24 (*Mdn*=16; *IQR*=5), and 24% participants scored ≥18, indicating significant levels of fatigue.

Mental Health (RCADS-25). Total anxiety T-scores ranged from 31 – 98 (*Mdn*=50.09; *IQR* =23.74), whilst total depression T-scores ranged from 37 – 87 (*Mdn*=52.06; *IQR*=12.20). The overall RCADS T-scores (combined anxiety and depression) ranged from 35 – 96 (*Mdn*=50.74; *IQR*=19.76), with 15% of participants scoring above threshold for probable clinical significance.

Sleep Quality (ASWS-S). Total mean score was 3.73 (*SD*=.76, range 2 - 5).

Physical Functioning (SF-36 PFS). Scores ranged from 0 – 95 (*Mdn*=45; *IQR*=65), indicating huge variation in self-reported physical health of participants.

Social Functioning (SSAS). This was only completed by 26 participants. Median score was 12.5 (*IQR*=15.25, range 2 - 32).

Health-Related QOL (KIdscreen-27). Total mean score was 89.82 (*SD*=14.16). Domains of physical (*M*=36.22; *SD*=6.14) and psychological (*Mdn*=35.49; *IQR*=10.06)

wellbeing, based on T-Scores, were rated lower compared to that of relationships with parents (*Mdn*=45.90; *IQR*=11.13), peers (*M*= 42.87; *SD*=11.84), and school (*Mdn*=45.38; *IQR*=8.13).

Cognitive and Behavioural Responses to Symptoms (CBRQ-S). Overall mean score was 32.55 (*SD*=11.81, range 8 - 55). Cognitive responses, including fear avoidance, embarrassment avoidance, damage beliefs, and symptom focusing, were rated higher than behavioural responses. See Table 3.2 for full descriptive information of each outcome measure.

Table 3.2

Descriptive Information from Outcome Measures

Measure	Full Sample					Interviewed participants				
	n	Mean (SD)	Mdn (IQR)	Range	%	n	Mean (SD)	Mdn (IQR)	Range	%
Fatigue (CFQ)	33	15.24 (4.21)*	16.00 (5)	2 - 24		12	15.92 (3.15)*	16.00 (4)	12-24	
Above threshold (≥18)	8				24.2	1				8.3
Below threshold (<18)	25				75.8	11				91.7
Combined anxiety and depression (RCADS-25)										
Total score	33	56.11 (15.65)*	50.64 (19.76)	35.36-96.13		12	58.92 (14.84)	56.98 (20.64)	42.18-93.37	
Normal range	25				75.8	8				66.7
Borderline threshold	3				9.1	2				16.7
Clinical threshold	5				15.2	2				16.7
Anxiety (RCADS-25)	33	54.51 (15.79)*	50.09 (23.74)	31.57-98.70		12	57.27 (13.94)	51.25 (21.54)	40.50-87.13	
Normal range	24				72.7	8				66.7
Borderline threshold	5				15.2	3				25.0
Clinical threshold	4				12.1	1				8.3
Depression (RCADS-25)	33	56.19 (12.13)*	52.06 (12.20)	37.49-87.08		12	57.93 (12.57)	55.20 (16.34)	37.49-85.15	
Normal range	27				81.8	9				75.0
Borderline threshold	1				3.0	1				8.3

Clinical threshold	5				15.2	2				16.7
Sleep quality (ASWS-S) Total Score	33	3.73 (0.76)	3.80 (1)	2 - 5		12	3.69 (0.73)	3.60 (1)	2 – 5	
Physical Functioning (SF36PFS)	33	43.64 (32.63)*	45.00 (65)	0 – 95		12	39.58 (32.71)	40.00 (70)	0 - 90	
Social functioning (SSAS)	26	13.50 (9.23)*	12.50 (15.25)	2 – 32		11	13.64 (5.94)	14.00 (11)	7 - 23	
HRQOL (Kidscreen-27)										
Physical	30	36.22 (6.14)	34.65 (8.28)	25.07-49.63		10	35.19 (5.88)	34.65 (9.42)	25.07-42.53	
Psychological	30	37.74 (5.73)*	35.49 (10.06)	29.42-50.61		10	35.19 (3.23)	34.32 (4.42)	31.96-41.75	
Parental	30	48.10 (9.97)*	45.90 (11.13)	35.96-74.39		10	43.65 (5.52)	42.87 (5.89)	35.96-55.75	
Peers	30	42.87 (11.84)	41.01 (11.11)	11.24-66.34		10	40.30 (8.16)	39.92 (13.45)	26.73-49.79	
School	28	46.19 (7.57)*	45.38 (8.13)	32.79-71.00		9	45.39 (10.79)*	42.94 (8.53)	32.79-71.00	
HRQOL total	28	89.82 (14.16)	89.00 (21)	62 – 124		9	85.67 (7.91)	83.00 (12)	75 – 100	
Cognitive and behavioural responses to symptoms (CBRQ-S)										
Total	33	32.55 (11.81)	32.00 (16)	8 - 55		12	37.17 (7.96)	38.00 (13)	26 – 50	
Fear avoidance	33	6.09 (2.94)	7.00 (4)	0 – 11		12	6.58 (2.88)	7.00 (5)	2 – 11	
Embarrassment avoidance	33	6.24 (3.87)	7.00 (7)	0 – 12		12	6.58 (3.48)	7.50 (6)	1 – 12	
Damage beliefs	33	6.45 (2.64)	7.00 (5)	0 – 11		12	7.25 (2.56)	7.50 (4)	2 – 11	
Symptom focusing	33	6.97 (2.47)	7.00 (4)	0 – 12		12	8.00 (1.95)	7.00 (2)	5 – 12	
All-or-nothing behaviour	33	3.94 (3.43)*	3.00 (6)	0 – 11		12	5.33 (3.68)	4.50 (7)	0 – 11	
Avoidance / Resting behaviour	33	2.85 (2.94)*	2.00 (3)	0 – 12		12	3.42 (2.35)	4.00 (4)	0 – 7	

Note. SD, Standard Deviation; IQR, Interquartile range; CFQ, Chalder Fatigue Questionnaire; RCADS-25, Revised Children's Anxiety and Depression Scale (higher scores, higher symptom levels); ASWS-S, Adolescent Sleep-Wake Scale-Short Version (higher scores, better sleep); SF-36 PFS, Short-Form 36

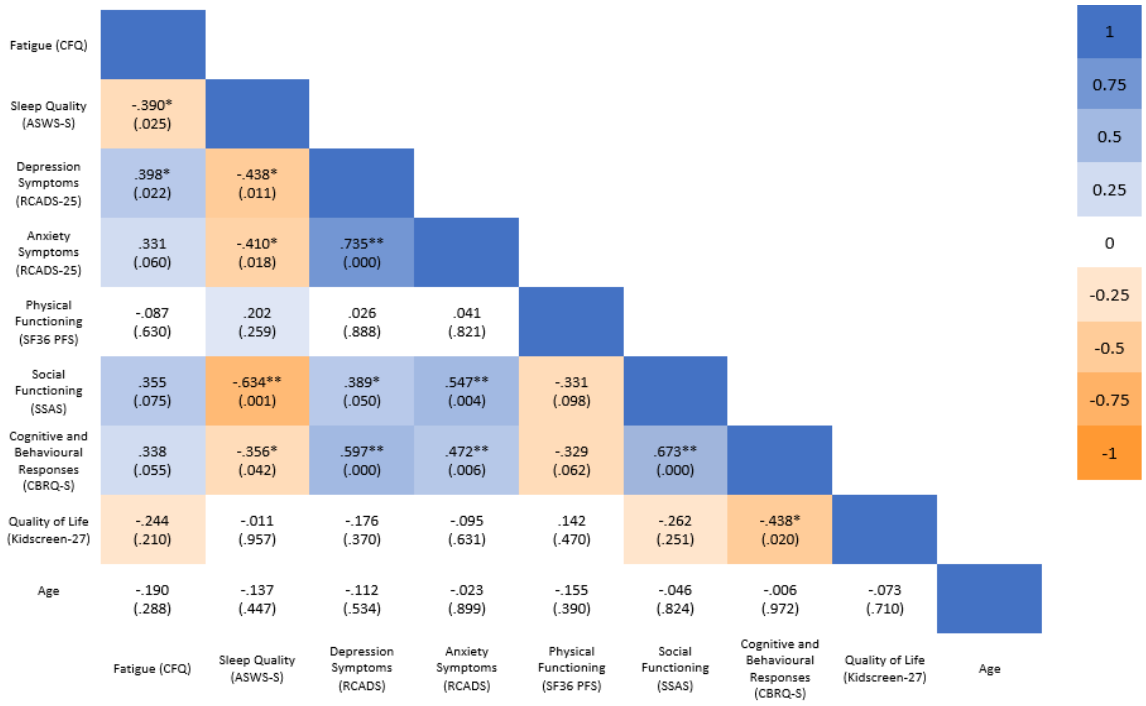
Physical Functioning Subscale (higher scores, better physical functioning); SSAS, School and Social Adjustment Scale (higher scores, more difficulties); CBRQ-S, Cognitive Behavioural Response Questionnaire – short version (higher scores, unhelpful responses); Kidscreen-27 (higher scores, better functioning). *Not normally distributed

Correlations

There was a moderate association (see Figure 3.2) between fatigue and sleep quality (*r* = -.39) and similarly between fatigue and depression (*r* =.40).

Figure 3.2

Correlation Analyses, Based on Spearman's Rank Correlation Coefficient, Between Levels of Fatigue and Variables of Interest



Note. CFQ, Chalder Fatigue Questionnaire; ASWS-S, Adolescent Sleep-Wake Scale-Short Version; RCADS-25, Revised Children's Anxiety and Depression Scale; SF-36 PFS, Short-Form 36 Physical Functioning Subscale; SSAS, School and Social Adjustment Scale; CBRQ-S, Cognitive Behavioural Response Questionnaire – short version
*p = <.05, ** p = <.001

Exploratory multiple stepwise linear regression (Table 3.3) found that depression symptoms explained 30% of the variance in fatigue, whilst sleep quality, anxiety, quality of life, social functioning, physical functioning, and cognitive and behavioural responses to symptoms did not significantly explain variance in fatigue.

Table 3.3

Exploratory Multiple Stepwise Linear Regression Analysis

Dependent variable	Predictor	β	p	B	SE	95% CI	R^2	Adjusted R^2
Fatigue (CFQ)	Depression (RCADS-25)	.549	.010	.191	.067	.051 - .330	.302	.265
	Age	-.105	.598					
	Gender	.015	.939					
	Sleep quality (ASWS-S)	-.089	.705					
	Anxiety (RCADS-25)	.130	.689					
	Physical functioning (SF36 PFS)	-.007	.974					
	Social functioning (SSAS)	.176	.444					
	Quality of life (Kidscreen-27)	-.198	.327					
	Cognitive and behavioural responses (CBRQ-S)	.054	.826					

Note: β, standardised coefficients; p indicates significance level; B, unstandardised coefficients; SE, standard error; 95% CI, 95% confidence interval, r^2 indicates percentage of variability in fatigue explained by predictors. CFQ, Chalder Fatigue Questionnaire; ASWS-S, Adolescent Sleep-Wake Scale-Short Version; RCADS-25, Revised Children's Anxiety and Depression Scale; SF-36 PFS, Short-Form 36 Physical Functioning Subscale; SSAS, School and Social Adjustment Scale; CBRQ-S, Cognitive Behavioural Response Questionnaire – short version

Qualitative Analysis (n=12)

Demographics and clinical characteristics of the subsample who were interviewed are outlined in Table 3.1 and Table 3.4.

Table 3.4

Demographic and Clinical Characteristics of the Subsample of Participants who were Interviewed

Participant	Condition	Gender	Age	Day-to-day	Experience of fatigue
1	Congenital myopathy	F	23	Unemployed	"Experienced [fatigue] for quite some time"
2	Myotonic dystrophy (type 1)	F	18	University student	"Sometimes… not always, I can get really tired in the day"
3	Charcot Marie tooth disease	M	22	University student	Yes "It is normal"
4	SMA (type 2)	F	24	Employed & student	Yes "I have … a battery for the day"
5	Becker muscular dystrophy	F	21	Self-employed	Yes "I experience it at a lot."
6	SMA (type 2)	F	24	Employed	Yes "I've always been tired"
7	Congenital myopathy	F	22	Volunteer	No (only in specific scenarios, e.g., illness, menstruation)
8	DMD	M	23	Employed	Yes "quite a lot"
9	FSHD	F	16	School student	Yes "I get tired pretty quickly"
10	FSHD	M	24	Employed	Yes "I can experience fatigue"
11	FSHD	F	23	Volunteer	Yes, linked with pain
12	Myotonic dystrophy (type 1)	F	22	College student	Yes "from time to time"

Note. SMA, Spinal muscular atrophy; DMD, Duchenne Muscular Dystrophy; FSHD, Facioscapulohumeral muscular dystrophy

Five overall themes were constructed (Table 3.5) which capture the lived experience of fatigue, including triggers, impact, the challenge of managing it, and internal and external responses. Detailed quotes are shown in Appendix J.

Table 3.5

Main Themes and Sub-themes Constructed from Interviews

Main Themes	Sub-themes
1. A constant uncertainty	- How fatigue was experienced - Degree of fatigue - Mind and body fatigue
2. Fatigue is multifactorial and can be a cycle	- Interconnected triggers for fatigue - Cycles between fatigue and the neuromuscular condition
3. Fatigue interferes with different areas of life	- Day-to-day tasks - Thinking abilities - Personal and social impact - Mood
4. It's tricky to manage fatigue	- Different things work at different times for different people - Changing plans - Push through fatigue
5. Mixed responses to fatigue (internal and external)	- Normalising - Minimising - Mixed feelings about disclosing fatigue and being treated differently - People try to be supportive but it's hard to understand - Fatigue is rarely discussed in healthcare

1. A constant uncertainty

How fatigue was experienced

Participants described fatigue as an extreme and "constant feeling of tiredness" (P6) and exhaustion. This experience was varied; for some, this feeling was "always" (P10) present and uncontrollable, causing participants to feel "tired all of the time" (P12). Whilst for others it happened "quickly" (P9). A common theme, however, was that it was unpredictable which made it difficult to plan for: "My bad day could be any day of the week, anytime. It could be tomorrow, and I wouldn't know." (P2).

Participants associated fatigue with reduced energy which made it difficult to continue activities. Some used analogies to explain this, likening fatigue to a draining battery or scale, for example: "Your energy levels are completely depleted… On a scale you've gone down to a zero." (P4).

Degree of fatigue

The degree to which participants experienced fatigue differed both between (see Table 4) and within participants. Different levels of fatigue severity were identified, from a feeling of tiredness that felt “manageable” (P10) to “absolute fatigue” (P4) that felt more intense and impacted health and day-to-day functioning.

> [There’s] just normal tiredness which is like constant and that's fine. And then there's fatigue, what I would deem as SMA [Spinal Muscular Atrophy] fatigue, it’s like my arms are dead, I can’t move them, I can't breathe properly, I want to cry the whole time… It’s like I just can’t function. (P6)

Mind and body fatigue

Some participants felt that fatigue affected them both physically and mentally:

> If I was going to put it in physical symptoms, it would be muscle aches, pains… as well as things like your brain going foggy, not being able to focus or find the right words to explain things. (P8)

Whilst others described a conflict between their physical state and mental aspirations, for example: “What your brain wants to do and what your body actually can allow you to do, it's a huge disparity that's very difficult to deal with” (P6).

2. Fatigue is multifactorial and can be a cycle

Interconnected triggers for fatigue

Many factors could exacerbate fatigue, including “stress” (P9), “cold” weather (P12), “driving” (P8), and hobbies. Participants described engaging in sports, for instance, but acknowledged that such hobbies often contributed to fatigue: “I do love it [sport activity] but… it gets me tired… I’ll be less likely to do it… if I'm tired that affects my motivation to do things I enjoy” (P2).

Paradoxically, inactivity could also contribute to fatigue: “If I'm just at home and I've not got really anything to do, I feel like it can affect me more… I'd feel more tired” (P7).

Participants also recognised connections between fatigue and sleep, whereby fatigue affects health, which disrupts sleep, exacerbating fatigue:

> It's a bit of a self-fulfilling cycle because if I'm tired, I then don't breathe very well over night and if I don't breathe very well over night, I don't sleep properly, which then means I'm more tired in the morning and then it just piles on. (P6)

Further cycles were created through short-term fixes to ease fatigue, such as caffeine, which further disrupted sleep and increased fatigue.

> When I'm tired, I get into bed, I can't sleep because obviously you've had all this caffeine … I spend all day trying to get into it, feeling sluggish … and then obviously soon as the caffeine hits it's like 9:00 o'clock and you just want to go sleep at that point. (P8)

Cycles between fatigue and the neuromuscular condition

Many viewed fatigue as central to their neuromuscular condition and part of "the weakness" in muscles (P1). Managing symptoms of the condition, such as pain or muscle weakness, required energy and increased fatigue, which could also exacerbate symptoms.

> I'll feel incredibly exhausted for a couple of days, and then I'll realise that I'm actually [in] quite a low level of pain, but a constant… dull… pain… Dealing with that can be quite exhausting and make me very fatigued and have zero energy. (P11)

3. Fatigue interferes with different areas of life

Day-to-day tasks

Participants described that fatigue "interferes a lot" (P5) with daily life and could "limit" (P4) abilities and activities which affects work, study, and livelihood.

Thinking abilities

Fatigue affected participants' thinking abilities, including concentration and memory which, in turn, further affected participants ability to work and study. Participants described brain "fog" (P8) which made it difficult to retain information, for example P9 described: "I won't be able to focus because I'm so tired. I just kind of sit there and think well there's no point in trying… if I can't remember anything."

Personal and social impact

Fatigue affected participants' personal and social lives, including self-esteem and independence. Participants identified negative automatic thoughts linked with fatigue,

such as feeling “useless” (P11) or “like an idiot” (P8) and wondered whether other people might think they were “incapable” (P8) as they might require more “help with things” (P4) when feeling fatigued.

Participants’ social life was also restricted as school and work often exacerbated fatigue and meant that weekday and evening activities were “off limits” (P8) as participants felt they needed to “reserve” (P4) energy for the following day as otherwise “work suffers” (P6). Similarly, fatigue left participants feeling isolated as they did not want to “see anybody” (P1) and wanted to avoid social interactions. Given the unpredictable nature of fatigue, participants also noted the need to cancel plans to prioritise rest: “you’re more exhausted… and I have no choice, I have to call off the event.” (P3)

Mood

The impact of fatigue on social life and hobbies had a clear interaction with participants’ mood as all noted feeling “easily” frustrated (P10) or low in mood related to missing out “on a lot” (P5) or feeling limited in activities.

> I have in the past been quite low, because I've not had the energy to do the tasks I've wanted to do… it made me feel quite angry with my own body, which is so bizarre because I've got no control over it. (P4)

Managing fatigue, alongside neuromuscular symptoms, caused participants to feel “different” (P5) to peers which also contributed to low mood. Participant 6 noted, for example, that fatigue “limits how much I can do, it limits the quality of how much I can do, and it’s a reminder of your disability which [affects] mental health”.

4. It’s tricky to manage fatigue

Different things work at different times for different people

It was challenging to manage fatigue and some reported that “nothing you do helps” (P12). There were inconsistent reports of whether rest or sleep was helpful in easing fatigue, and this contributed to the idea that different things helped at different times, which “depends” (P4) on fatigue levels. Others noticed feeling preoccupied with fatigue and wanting to rest but noted that this was not always helpful as it can “make you even more tired” (P2).

Further inconsistencies were reported as rest or periods of downtime exacerbated fatigue or made it more noticeable, for example “I have those moments of … down period

where it just hits me." (P12). Paradoxically, participants also reported that keeping active, whilst a possible trigger for fatigue (Theme 2), could "make it a little easier" (P3).

Changing plans

Participants reported needing to "stick to a schedule" (P4) and prioritise tasks to conserve energy throughout the day. This required more thought and "mental calculation" (P6) which may have further contributed to mental fatigue.

Push through

Participants also reported trying to persevere with activities or "push through" (P12) despite fatigue. Whilst that could help "get through the day" (P10) or engage in enjoyable activities, participants often found that this had consequences on recovery time and on activities the following day.

5. Mixed responses to fatigue (internal and external)

Normalising

Participants viewed fatigue as "normal" (P1) and something they were "used to" (P12). They also saw it as normal within the neuromuscular community: "I know it's quite a common thing, with … conditions … to be fatigued" (P7), and perhaps as a "normal" (P3) feeling for everyone: "I can experience fatigue, but I don't know if this fatigue is generally normal in people or it's something like in myself" (P10).

Minimising

Alongside normalising attitudes, participants seemed to minimise the effects of fatigue. Whilst participants described wide-ranging consequences, and CFQ scores indicate the presence of fatigue, they did not always feel that there was an impact of fatigue, for example: "Sometimes I do nothing all day because I get really tired, but fatigue doesn't really affect me too much" (P2).

Mixed feelings about disclosing fatigue and being treated differently

Participants reported mixed feelings about discussing fatigue with friends, family, and employers. Some preferred to manage it "alone" (P1) as they did not "want to be treated any differently" (P2). Others approached fatigue with a sense of normality and described that it was "not a big enough issue to discuss with anyone" (P10). In contrast, some participants described reasonable adjustments and benefits of being treated

differently, for example P9 indicated that teachers reduced their expectations “during the last lessons” due to fatigue levels.

People try to be supportive but it’s hard to understand

Those that did rely on social support to help manage fatigue reported that it was difficult to “explain to other people” (P12), and it was not always possible for others to understand. This led to mixed responses or questions, such as “is it even real?” (P5) and meant that friends, family, and employers sometimes got it wrong in terms of the support offered, for example offering an “easy solution” (P8) or making assumptions “that everyone is the same” (P7) and would benefit from the same support.

Despite this, participants reported that social support was also beneficial at times as others were better able to “recognise” (P11) fatigue, offer “help” (P4) and encourage gentle “activity” (P3). One participant noted the difficult balance of relying on others whilst maintaining independence:

> If there's a task that somebody else can complete for me in much less time and it has no effect on them, I would rather them do it … I know that can be quite difficult for people, but … as I've got older, I've appreciated the balance of not losing my independence, but not overexerting myself. (P4)

Fatigue is rarely discussed in healthcare

Participants reported that fatigue was “rarely” (P8) discussed by healthcare professionals and was often overlooked or “ignored” (P4). When participants did discuss fatigue, they found that this was often thought about in terms of “managing pain” (P11) or medical features, rather than “the impact” (P4) of fatigue.

Participants reported that it “would help a lot” (P5) for clinicians to discuss fatigue, to “have it recognised as a symptom” (P6) and offer “tools or things I could do to help me maintain my energy” (P4).

Discussion

We found that 24% of our self-selecting sample of young people with NMD experienced significant levels of fatigue. This indicates that at least some level of fatigue is common for young people across neuromuscular conditions and has a wide-ranging impact on daily functioning. Fatigue was linked to sleep quality and depression, both in our quantitative data and qualitative descriptions elicited through interviews. Interviews

captured the lived experience of physical and mental fatigue, potential triggers, the impact, challenges of managing it, and internal and external responses to fatigue.

Fatigue is a common experience for adults with neuromuscular conditions (Pangalila et al., 2015) and this study indicates that this is similar for young people. Although our opt-in approach to recruitment makes it difficult to draw conclusive prevalence estimates, our findings are consistent with previous research which has reported similar prevalence rates of fatigue in young people with specific neuromuscular conditions, such as Charcot-Marie-Tooth disease (Jagersma et al., 2013) and in those with other chronic health conditions, such as HIV (Coetzee et al., 2018). Qualitative data highlighted the paradoxical experience of fatigue as ideas used to ease fatigue, such as either keeping active or resting, could also trigger feelings of fatigue. Our participants also described varying levels of fatigue, with mental and physical effects, which is similar to descriptions noted by young people with chronic fatigue syndrome (Parslow et al., 2018) and other chronic health conditions (Poku et al., 2020). This indicates that fatigue may be a transdiagnostic factor across neuromuscular conditions, as well as chronic health conditions more generally.

Of the clinical characteristics explored within this study, symptoms of depression and sleep quality showed strongest associations with fatigue. This is consistent with findings from previous research (El-Aloul et al., 2020) with young males with DMD. The subsample of participants who were interviewed also reported links between sleep and fatigue, with some participants identifying cycles between fatigue affecting health, health disrupting sleep, and exacerbating fatigue. Significant associations between fatigue and physical health, however, were not noted in the quantitative data. This could reflect the wide variation in self-reported physical functioning which is expected given the heterogeneity of neuromuscular conditions that affect individuals to varying levels. The inclusion of objective measures for physical functioning may have helped to further identify patterns.

Despite physical functioning not showing a significant relationship with fatigue, interview participants did report links between pain and muscle weakness which they felt contributed to fatigue levels. This is consistent with a model of fatigue developed with adults with NMD (Kalkman et al., 2007), however psychological factors, which are not considered within this model, were a key finding of our study with depression symptoms explaining 30% of fatigue variance. Interviewed participants noted the impact of fatigue and energy levels on mood as they felt different to peers, limited in their activities, and at times reported socially isolating themselves to cope with fatigue; this may further

exacerbate fatigue and low mood due to inactivity. This is similar to qualitative findings in young people with low mood and chronic fatigue syndrome (Taylor et al., 2017). These findings also align with the cognitive behavioural model of fatigue (Surawy et al., 1995) which recognises the biological, environmental and psychological contributors to fatigue.

Measures of HRQOL showed that individual domains, such as psychological and physical wellbeing, were rated lower than domains exploring external relationships with parents, peers, and school. These factors, however, did not show a significant relationship with fatigue, despite the subsample of interviewed participants reporting that fatigue limits quality of activities. Our qualitative data is consistent with previous research within paediatric neuromuscular conditions in which quality of life is associated with fatigue (Bhullar et al., 2018; Jagersma et al., 2013; Wei et al., 2016). A systematic review (Travlos et al., 2017) of non-ambulant young people with NMD found that psychosocial quality of life ratings were mixed, with younger adolescents with DMD and progressive trajectories showing reduced wellbeing scores compared to older adolescents. A wide range of demographic and clinical characteristics were included in this study, with participants being both ambulant and non-ambulant and ranging in age from 12-24; such variation in age and trajectory may help to explain inconsistencies between the quantitative data and qualitative findings and that of other studies.

Similarly, whilst participants described the impact of fatigue on social functioning, a significant correlation was not found in quantitative data. This measure may have been impacted due to a reduced completion rate. Interviewed participants also reported trying to “push through” fatigue and continue social activities despite fatigue levels, this coupled with minimising the impact of fatigue, may explain why social difficulties were not noted within quantitative measures and highlights the value of the qualitative descriptions to contextualise the findings. The “push through” attitude is well-described within fatigue literature and contributes to ‘boom and bust’ maintenance cycles of fatigue (Loades et al., 2019). Interestingly, this cycle was again not captured through quantitative measures (CBRQ-S) which looked at all-or-nothing behavioural responses to symptoms; it may be that these self-report measures need to be adapted to be more appropriate and relevant to young people with NMD, or that new measures need to be developed to capture the unique difficulties experienced by this group specifically.

Limitations

Recruitment relied on a self-selection process and so it is possible that those who took part were more likely to have experiences of fatigue. The prevalence estimates may therefore not be generalisable to young people with NMD more broadly but does suggest

that fatigue is common within this population. A further limitation is the use of the SSAS questionnaire to measure social functioning which includes items focused on school-based social activities and showed a reduced rate of completion amongst participants. The initial age range for this study focused on school-aged adolescents (age 11-18) but, due to difficulties with recruitment, was extended to capture those aged between 10 and 24 years, based on the World Health Organisation (2014) definition of young people. Those who were not school-aged may have felt that this questionnaire was not relevant to them, and this may have impacted the findings associated with social functioning.

Conclusions are further limited due to an underpowered sample size, however as participants experienced rare and progressive chronic health conditions, a large sample size was not expected. The addition of qualitative interviews ensure that this research is still valuable, particularly as previous research with this population has been limited. To our knowledge, this is the first mixed-method study regarding fatigue for young people with different NMDs. Findings are therefore helpful to inform services and possible psychological interventions for this population.

Clinical and research Implications

Our findings indicate that fatigue is a common concern for this population and appears to be linked to mood. Whilst larger, more representative samples are needed, our findings suggest that fatigue should be routinely screened within clinical settings for young people with NMDs, particularly when depression symptoms are present. Promising interventions have been identified (Higson-Sweeney et al., Submitted) for young people with fatigue, including CBT, which could be beneficial for young people with NMD due to links between fatigue and depression and negative automatic thoughts identified in interviews. Future research should explore the use and efficacy of psychological interventions for this population.

Conclusion

In conclusion, fatigue is a common experience for young people with NMDs, with varying levels of impact on day-to-day functioning and mood, which can be difficult for individuals to manage. This study indicates that fatigue is linked to quality of sleep and symptoms of depression. Responses to fatigue and ways in which it impacts on young people's physical and social functioning may not necessarily be detected using self-report questionnaires but was evident from their narratives.

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Executive Summary

Literature Review

Those with chronic health conditions often report fatigue as a common experience, with research showing that fatigue can affect physical and mental health as well as many areas of life. Whilst studies have explored fatigue within children and young people with specific chronic health conditions, research has not previously collated prevalence information across conditions. This review aimed to systematically synthesise the existing literature on the prevalence of fatigue within children and young people living with chronic health conditions. Given heterogeneity in the studies, a narrative synthesis approach was used instead of meta-analysis.

Three databases were searched to identify studies from January 2000 to July 2021. A total of 6875 papers were identified; the title and abstract of each paper was reviewed based on inclusion and exclusion criteria, and the full text was assessed of those that were potentially relevant. After removing duplicates, applying the eligibility criteria, and hand-searching the reference list of included papers, a total of 26 papers (25 studies) were included.

The papers comprised of a total of 2541 participants, ranging in age from 2 to 22 years, and included 12 categories of chronic health condition. The quality of studies was assessed using an adapted version of the Newcastle-Ottawa Scale and 13 were shown to be of high quality. Whilst findings showed that the prevalence of fatigue varied widely, within and between studies, some level of fatigue was common across conditions. Discrepancies were noted between child and parental reports, with parental ratings being an average of 26% higher than children and young people reported themselves. Studies that included comparisons with healthy peers indicated that prevalence and levels of fatigue tended to be higher in those with chronic health conditions. This suggests that it may be useful for services to routinely screen for fatigue when working with young people with chronic health conditions and to gather information from multiple sources. These implications along with recommendations for future research are discussed.

Service-Related Project

Research and the clinical use of mindfulness has grown substantially, with many clinical interventions and services incorporating mindfulness-based skills and exercises to help individuals develop an awareness and acceptance of the present moment. Previous

literature shows that learning mindfulness-based skills can reduce psychological distress, improve quality of life, and that group interventions can be effective for working-age adults and older adults.

Between 2015 and 2020, an NHS secondary mental health service has run a 12-week ‘Mindful Life’ group for clients aged 18 and over, with a range of diagnoses. A previous project had interviewed some participants of the group who had found the intervention to be beneficial, however whilst the service had also routinely collected outcome measures of psychological distress and mindfulness, these had not been evaluated. This service-related project aimed to evaluate the outcomes of this group, with specific focus on the following research questions: Over the course of the group, (1) is there a reduction in symptoms of distress; (2) is there an increase in mindfulness-based skills; (3) is there an association between overall mindfulness ratings and distress; (4) are there particular areas of mindfulness associated with more positive outcomes?

Over the five years, 201 people had been referred to the group with a total of 14 cohorts. Whilst 111 people were invited to attend the group, 70 participants completed the group, and 50 completed outcome measures at the start and end of the intervention. The majority of participants had diagnoses of anxiety or mood disorders. Analyses found a slight, but non-significant, decrease in levels of overall distress from the start to end of the group intervention and a significant increase in overall mindfulness-based skills. No significant correlations were found between overall mindfulness and overall distress levels; however, one specific area of mindfulness which involves not reacting to inner experience did demonstrate a significant negative correlation with distress. This indicates that as this specific skill increases, distress decreases. The findings were developed into recommendations and shared with the group facilitators.

Main Research Project

Neuromuscular disorders (NMD) are chronic and progressive conditions associated with muscle weakness which can lead to health complications and mobility difficulties. Fatigue has been reported to affect adults with NMD and is well documented in young people with other chronic health conditions. Whilst recent research has started to explore fatigue within young people with specific types of NMD, such as Duchenne Muscular Dystrophy which only affects males, fatigue has not been explored within those with other types of neuromuscular conditions. The current study aimed to contribute to the literature by investigating the prevalence, severity, and impact of fatigue amongst young

people with a range of neuromuscular conditions, and to explore correlates of fatigue for this population.

A cross-sectional mixed methods study was carried out; participants (*n*=33) completed seven online questionnaires regarding fatigue, mood, sleep, physical health, quality of life, social functioning, and cognitive and behavioural responses to symptoms. Participants ranged in age from 12 to 24 years and ten neuromuscular conditions were reported amongst participants. A subsample of participants (*n*=12) completed an online semi-structured interview regarding their experiences of fatigue which was analysed with Reflexive Thematic Analysis.

Overall, 24% of participants reported significant levels of fatigue, with correlations noted between fatigue and both sleep quality and symptoms of depression. Exploratory multiple stepwise linear regressions showed that depression explained 30% of the variance of fatigue which indicates that clinicians should screen for fatigue when working with this population, particularly when depression is reported. Five themes were constructed from qualitative interviews, which reflected the lived experience of fatigue, interconnected cycles, the far-reaching impact, internal and external responses to fatigue, and the challenges of managing it. Whilst further investigation and larger samples are needed, these findings indicate that fatigue is a common experience for young people with NMD and has a varied impact on day-to-day life. The findings are discussed in terms of the clinical implications and recommendations for future research.

Acknowledgements

Thank you to my clinical tutor, Dr Pamela Jacobsen, for keeping me on track throughout the three years and to my research supervisor, Dr Maria Loades, not only for your research guidance, but for being more organised than I could ever hope to be and for your calming presence when I have felt overwhelmed. I am also grateful for the support from Dr Anna Strudwick for my service-related project, and thankful to Dr Hen Joannidi, and Michael Houser for introducing me to the Mindful Life group, and to Dr Sadie Thomas-Unsworth, without whom there would have been no starting ideas for my main project.

Thanks to all those who have helped with my research along the way: Cath Borwick, for support with search terms; Dr Kate Chapple, for providing a clinical perspective; Nina Higson-Sweeney, for helping me understand systematic reviews; Paul Reynolds-Cowie and Srushti Gala for support with screening; the many incredible people working across charities and research registries that helped with advertising my main project; and those with lived experience who offered time and guidance. A special thanks to Ashleigh Westgate, an invaluable research apprentice, and to all those who took the time to participate in my research and shared your stories with me.

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On a personal note, I would like to thank my friends for offering helpful distractions and being my proof-readers; my mum for her thoughtfulness and optimism; my dad for his confidence; and a special thank you to my partner, Oli, who has been with me from day one of my first Assistant Psychologist role, supported my DClinPsy applications, and moved across the country with me. Thank you for being my personal cheerleader and, whilst not a psychologist, often a better psychologist than me. Thank you for being creative when I struggled to be and for talking me through grounding exercises when I most needed it.

Finally, I would like to thank Cohort 2019; although the pandemic isolated us at times and we missed out on a lot of time together, I am so grateful to have shared this journey with you all and have appreciated the support, cake, and peer supervision.

Appendices

Appendix A

Literature Review Author Guidelines – Journal of Psychosomatic Research

The *Journal of Psychosomatic Research* is a multidisciplinary research journal covering all aspects of the relationships between **psychology** and **medicine**. The scope is broad and ranges from basic human biological and psychological research to evaluations of **treatment** and services. Papers will normally be concerned with **illness** or **patients** rather than studies of healthy populations. Studies concerning special populations, such as the elderly and children and adolescents, are welcome. In addition to peer-reviewed original papers, the journal publishes editorials, reviews, and other papers related to the journal's aims.

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Introduction

Types of article

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It is not necessary to include detailed descriptions on the program or type of grants and awards. When funding is from a block grant or other resources available to a university,

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Example: '..... as demonstrated [3,6]. Barnaby and Jones [8] obtained a different result ....'
List: Number the references (numbers in square brackets) in the list in the order in which they appear in the text.
Examples:
Reference to a journal publication:
[1] J. van der Geer, J.A.J. Hanraads, R.A. Lupton, The art of writing a scientific article, J. Sci. Commun. 163 (2010) 51–59. https://doi.org/10.1016/j.Sc.2010.00372.
Reference to a journal publication with an article number:
[2] J. van der Geer, J.A.J. Hanraads, R.A. Lupton, 2018. The art of writing a scientific article. Heliyon. 19, e00205. https://doi.org/10.1016/j.heliyon.2018.e00205.
Reference to a book:
[3] W. Strunk Jr., E.B. White, The Elements of Style, fourth ed., Longman, New York, 2000.
Reference to a chapter in an edited book:
[4] G.R. Mettam, L.B. Adams, How to prepare an electronic version of your article, in: B.S. Jones, R.Z. Smith (Eds.), Introduction to the Electronic Age, E-Publishing Inc., New York, 2009, pp. 281–304.
Reference to a website:
[5] Cancer Research UK, Cancer statistics reports for the UK. http://www.cancerresearchuk.org/aboutcancer/statistics/cancerstatsreport/, 2003 (accessed 13 March 2003).

Reference to a dataset:
[dataset] [6] M. Oguro, S. Imahiro, S. Saito, T. Nakashizuka, Mortality data for Japanese oak wilt disease and surrounding forest compositions, Mendeley Data, v1, 2015. https://doi.org/10.17632/xwj98nb39r.1.
Reference to software:
[7] E. Coon, M. Berndt, A. Jan, D. Svyatsky, A. Atchley, E. Kikinzon, D. Harp, G. Manzini, E. Shelef, K. Lipnikov, R. Garimella, C. Xu, D. Moulton, S. Karra, S. Painter, E. Jafarov, S. Molins, Advanced Terrestrial Simulator (ATS) v0.88 (Version 0.88), Zenodo, March 25, 2020. https://doi.org/10.5281/zenodo.3727209.

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Appendix B

Literature Review Search Strategy

PsycNET

Filters:

Date Range: 2000 – 2021

Age group: Childhood (birth to 12 years), neonatal, infancy, preschool age, school age, adolescence (13 to 17 yrs), young adulthood (18 to 22 yrs)

Population group: humans

#1	Any Field: Adolescen* OR Any Field: Child* OR Any Field: "Young pe*" OR Any Field: Teen* OR Any Field: Juvenile* OR Any Field: Youth* OR Any Field: Minor* OR Any Field: Puberty OR Any Field: Pubescent* OR Any Field: Prepubescent* OR Any Field: School* OR Any Field: "High school*" OR Any Field: "High-school*" OR Any Field: "Secondary school*" OR Any Field: "School child*" OR Any Field: "Schoolchild*" OR Any Field: "School-child*" OR Any Field: "Primary school" OR Any Field "Primary-school" OR Any Field: Paediatric* OR Any Field: Pediatric* OR Any Field: Boy* OR Any Field: Girl*
#2	Index Terms: Fatigue OR Any Field: fatigue* OR Any Field: "Lack of energy" OR Any Field: "Loss of energy" OR Any Field: "Low energy" OR Any Field: Tired* OR Any Field: Drowsy OR Any Field: Drowsiness OR Any Field: Exhausted OR Any Field: Exhaustion OR Any Field: Lethargy OR Any Field: Lethargic OR Any Field: Weary OR Any Field: Weariness OR Any Field: Drained OR Any Field: Sleepy OR Any Field: Sleepiness OR Any Field: Lassitude OR Any Field: Sluggish* OR Any Field: Apath* OR Any Field: Astheni* OR Any Field: Malaise
#3	1 and 2
#4	Index Terms: "Chronic Illness" OR Any Field: "Chronic disease*" OR Any Field: "Chronic condition*" OR Any Field: "Chronic disorder*" OR Any Field: "Chronic illness*" OR Any Field: "Chronic syndrome*" OR Any Field: "Chronic health" OR Any Field: "Long-term condition*" OR Any Field: "Long-term disease*" OR Any Field: "Long-term disorder*" OR Any Field: "Long-term illness*" OR Any Field: "Long-term syndrome*" OR Any Field: "Long term condition*" OR Any Field: "Long term disease*" OR Any Field: "Long term disorder*" OR Any Field: "Long term illness*" OR Any Field: "Long term syndrome*" OR Any Field: "Longstanding Condition*" OR Any Field: "Longstanding disease*" OR Any Field: "Longstanding illness*" OR Any Field: "Longstanding syndrome*" OR Any Field: "Longstanding disorder*" OR Any Field: "Long-standing condition*" OR Any Field: "Long-standing disease*" OR Any Field: "Long-standing disorder*" OR Any Field: "Long-standing illness*" OR Any Field: "Long-standing syndrome*"
#5	Index Terms: AIDS OR Any Field: AIDS OR Any Field: "Acquired immunodeficiency syndrome"
#6	Any Field: "Allergic Rhinitis" OR Any Field: "Hay fever" OR Any Field: Hay-fever OR Index Terms: "Hay Fever" OR Index Terms: "Respiratory Tract Disorders"
#7	Index Terms: "Allergic disorders" OR Any Field: Allerg* OR Index Terms: "Allergic Skin Disorders"
#8	Index Terms: Anemia OR Index Terms: "Blood and Lymphatic Disorders" OR Index Terms: "Genetic Disorders" OR Any Field: Anaem* OR Any Field: "Fanconi Anaemia" OR Any Field: Anem* OR Any Field: "Fanconi Anemia"
#9	Index Terms: "Angina Pectoris" OR Any Field: Angina OR Index Term: "Heart Disorders"

#10	Index Terms: “Arrhythmias (Heart)” OR Any Field Arrhythm*
#11	Index Terms: Arthritis OR Index Terms: “Rheumatoid Arthritis” OR Any Field: Arthriti* OR Any Field “Juvenile Arthritis”
#12	Index Terms: Asthma OR Any Field Asthma*
#13	Index Terms: “Fibrillation (Heart)” OR Any Field: “Atrial fibrillation”
#14	Index Terms: “Immunologic Disorders” OR Any Field “Autoimmune Disease” OR Any Field: “Autoimmune Diseases”
#15	Index Terms: Neoplasms OR Any Field: Cancer* OR Any Field Oncolog* OR Index Terms: Oncology
#16	Index Terms: “Cerebral Palsy” OR Any Field: “Cerebral Palsy” OR Index Terms: “Brain Disorders” OR Index Terms: “Central Nervous System Disorders”
#17	Any Field: Gout OR Any Field: Gouty OR Any Field: “Gouty Arthritis”
#18	Index Terms: “Kidney Diseases” OR Any Field: “Kidney disease” OR Any Field: “Kidney Diseases” OR Any Field: “Kidney failure”
#19	Index Terms: “Chronic Obstructive Pulmonary Disease” OR Any Field: COPD OR Any Field: “Chronic Pulmonary Disease” OR Index Terms: “Lung Disorders”
#20	Index Terms: “Chronic Pain” OR Any Field: “Chronic pain”
#21	Any Field: Sarcoidosis OR Any Field: sarcoid*
#22	Index Terms: “Cleft Palate” OR Index Terms: “Congenital Disorders” OR Index Terms: “Neonatal Disorders” OR Any Field: “Cleft Lip” OR Any Field: “Cleft Palate”
#23	Any Field: “Coeliac disease” OR Any Field: Coeliac* OR Any Field: “Celiac disease” OR Any Field: Celiac* OR Index Terms: “Celiac Disease”
#24	Any Field: “Connective tissue disease” OR Any Field: “Connective tissue diseases” OR Any Field: Lupus OR Index Terms: Lupus
#25	Index Terms: “Heart Disorders” OR Index Terms: “Cardiovascular Disorders” OR Any Field: “Coronary Heart Disease” OR Any Field: “Heart Disease” OR Any Field: “Heart Diseases”
#26	Index Terms: “Colon Disorders” OR Index Terms: “Gastrointestinal disorders” OR Any Field: crohn’s OR Index Terms: “Ulcerative colitis” OR Any Field: “inflammatory bowel disease” OR Any Field: IBD
#27	Index Terms: “Cystic Fibrosis” OR Any Field: “Cystic Fibrosis” OR Any Field*:* CF
#28	Index Terms: Diabetes OR Index Terms: “Diabetes Mellitus” OR Any Field: Diabet*
#29	Index Terms: Eczema OR Index Terms: Dermatitis OR Any Field: Eczema*
#30	Any Field: Endocrine OR Any Field: “Endocrine Diseases” OR Any Field: “Endocrine disease” OR Any Field: Thyroid OR Any Field: Adrenal OR Index Terms: “Endocrine System” OR Index Terms: “Endocrine disorders”
#31	Any Field: Endometri*
#32	Index Terms: Epilepsy OR Any Field: Epilep* OR Index Terms: “Epileptic Seizures” OR Index Terms: Seizures
#33	Index Terms: Fibromyalgia OR Index Terms: “Muscular Disorders” OR Any Field: Fibromyalgia
#34	Index Terms: Hemophilia OR Any Field: Haemophil* OR Any Field: Hemophil*
#35	Any Field: “Heart failure” OR Any Field: “Heart condition*” OR Any Field: “Heart Disease*” OR Any Field: “Heart Defect*”
#36	Any Field: “Hepatitis B” OR Index Terms: Hepatitis OR Index Terms: “Toxic hepatitis” OR Index Terms: “Liver Disorders”
#37	Any Field: HIV OR Any Field: “Human Immunodeficiency Virus” OR Index Terms: HIV

#38	Any Field: Hypertens* OR Any Field: Hypotens* OR Any Field: “High Blood Pressure” OR Index Terms: Hypertension OR Index Terms: “Blood pressure disorders”
#39	Any Field: “Lung diseases” OR Any Field: “Lung disease” OR Any Field: “Lung disorder” OR Any Field: “Lung disorders” OR Any Field: “Lung fibrosis”
#40	Any Field: “Neurometabolic disorder*” OR Any Field: “Neurometabolic condition*” OR Any Field: “Neurometabolic disease*” OR Any Field: Metabolic OR Index Terms: “Metabolic Syndrome”
#41	Any Field: “Migraine-neuralgia” OR Any field: Migraine OR Index Terms: “Migraine Headache” OR Index Terms: “Muscle Contraction Headache” OR Index Terms: Headache
#42	Any Field: Neuralg* OR Index Terms: Neuralgia OR Index Terms: “Trigeminal neuralgia”
#43	Index Terms: “Neuromuscular Disorders” OR Index Terms: “Nervous System Disorders” OR Index Terms: “Muscular Dystrophy” OR Index Terms: “Myasthenia Gravis” OR Index Terms: “Muscular Disorders” OR Index Terms: “Musculoskeletal Disorders” OR Any Field: “Neuromuscular Disease*” OR Any Field: “Neuromuscular Condition*” OR Any Field: “Neuromuscular Disorder*”
#44	Any Field: "Chronic otitis media" OR Any Field: “Otitis media”
#45	Any Field: "Peripheral Arterial Disease" OR Any Field: "peripheral vascular disease" OR Any Field: PAD
#46	Any Field: Phenylketonuria OR Any Field: PKU OR Index Terms: Phenylketonuria OR Index Terms: “Metabolism Disorders”
#47	Any Field: "Polycystic ovary syndrome" OR Any Field: “polycystic ovaries” OR Any Field: PCOS
#48	Any Field: Psoriasis OR Index Terms: “Skin Disorders”
#49	Any Field: “Sickle cell” OR Index Terms: “Sickle cell disease”
#50	Any Field: Sjörgen* OR Any Field: Sjorgen
#51	Any Field: “Sleep disorder” OR Any Field: “Sleep disorders” OR Any Field: “Sleep apnoea” OR Index Terms: Narcolepsy OR Index Terms: “Sleep apnea” OR Index Terms: “Sleep wake disorders” OR Index Terms: Insomnia
#52	Any Field: “Spina bifida” OR Index Terms: “Spina bifida”
#53	Any Field: “Spinal cord” OR Any Field: “Spinal disease” OR Any Field: “Spinal diseases” OR Any Field: “Spinal injury” OR Any Field: “Spinal injuries” OR Index Terms: “Spinal cord injuries” OR Index Term: “Spinal cord”
#54	Any Field: Thalassemia OR Any Field: Thalassem*
#55	Any Field: Thyroid OR Any Field: Hyperthyroid* OR Any Field: Hypothyroid* OR Index Terms: Hypothyroidism OR Index Terms: Hyperthyroidism OR Index Terms: “Thyroid disorders“
#56	Combine searches 4 – 55 using OR
#57	Combine searches 3 AND 56
#58	Any field: prevalence OR any field: prevalen* OR any field: risk* OR any field: rate* OR any field: trend OR any field: associat*
#59	Combine searches #58 AND #57

EMBASE

Filters

Date range: 2000 – 2021

Population: humans, only in English, Sources: Embase, Embase Classic

Age filter: Newborn (0-1 month), infant (1-12 months), Child (1-12 years), Preschool child (1-6 years), School child (7-12 years), Adolescent (13-17 years), young adult (18-24 years).

/exp = exploded term (includes the databases identified synonyms)

#1	'adolescent'/mj OR 'child'/mj OR adolescen* OR child* OR 'young person' OR 'young people' OR teen* OR juvenile* OR youth* OR minor* OR puberty OR pubescent* OR prepubescent* OR school* OR 'high school' OR 'high-school' OR 'secondary school' OR 'school child*' OR 'schoolchild*' OR 'school-child*' OR paediatric* OR pediatric* OR boy* OR girl* OR 'primary school' OR 'primary-school'
#2	'fatigue'/exp OR fatigue* OR 'lack of energy' OR 'loss of energy' OR 'low energy' OR tired* OR drows* OR lethargy OR lethargic OR exhaust* OR weary OR weariness OR drained OR sleepy OR sleepiness OR lassitude OR sluggish* OR apathy OR apathetic OR astheni* OR malaise
#3	#1 AND #2
#4	'chronic disease'/exp OR 'chronic illness*' OR 'chronic condition*' OR 'chronic disorder*' OR 'chronic syndrome*' OR 'chronic health' OR 'long-term condition*' OR 'long-term disease*' OR 'long-term disorder*' OR 'long-term illness*' OR 'long-term syndrome*' OR 'long term condition*' OR 'long term disease*' OR 'long term disorder*' OR 'long term illness*' OR 'long term syndrome*' OR 'longstanding condition*' OR 'longstanding disease*' OR 'longstanding illness*' OR 'longstanding syndrome*' OR 'longstanding disorder*' OR 'long-standing condition*' OR 'long-standing disease*' OR 'long-standing disorder*' OR 'long-standing illness*' OR 'long-standing syndrome*'
#5	'Acquired immune deficiency syndrome'/exp OR 'acquired immune deficiency syndrome'
#6	'Allergic Rhinitis'/exp or 'allergic rhinitis'
#7	'allergy'/exp OR 'allergy' OR 'allergic disease'/exp OR 'allergic disease' OR 'allergic disorder*' OR 'allerg*' OR 'skin allergy'/exp OR 'skin allergy'
#8	'Anemia'/exp OR 'Anemia' *OR* 'Fanconi anemia'/exp *OR* 'Fanconi anemia' OR 'genetic disorder'/exp OR 'genetic disorder' OR 'anaem*' OR 'anem*'
#9	'Angina pectoris'/exp OR 'angina pectoris'
#10	'Heart arrhythmia'/exp OR 'heart arrhythmia'
#11	'Arthritis'/exp or 'Arthritis' *OR* 'Juvenile rheumatoid arthritis'/exp *OR* 'Juvenile rheumatoid arthritis'
#12	'Asthma'/exp OR 'Asthma'
#13	'Atrial fibrillation'/exp OR 'Atrial fibrillation'
#14	'Autoimmune disease'/exp OR 'Autoimmune disease' OR 'Immunopathology'/exp or 'Immunopathology'
#15	'Hematologic disease'/exp OR 'Hematologic disease'
#16	'Malignant neoplasm'/exp OR 'Neoplasm'/exp OR 'Malignant neoplasm' OR 'Neoplasm'
#17	'Cerebral palsy'/exp OR 'Brain disease'/exp OR 'Central nervous system disease'/exp OR 'Cerebral palsy' OR 'Brain disease' OR 'Central nervous system disease'/
#18	'Gout'/exp OR 'Gout'
#19	'Kidney disease'/exp OR 'Kidney disease'
#20	'Chronic obstructive lung disease'/exp OR 'Lung disease'/exp OR 'Chronic obstructive lung disease' OR 'Lung disease'

#21	‘Chronic pain’/exp OR ‘Chronic pain’
#22	‘Sarcoidosis’/exp OR ‘Sarcoidosis’
#23	'Cleft lip'/exp OR 'Cleft palate'/exp OR ‘Genetic disorder’/exp OR ‘Newborn disease’/exp OR 'Cleft lip' OR 'Cleft palate' OR ‘Genetic disorder’ OR ‘Newborn disease’
#24	‘Celiac disease’/exp OR ‘Coeliac’ OR ‘Celiac’ OR ‘Celiac disease’
#25	‘Connective tissue disease’/exp OR ‘Lupus erythematosus’/exp OR ‘Lupus Vulgaris’/exp OR ‘Connective tissue disease’ OR ‘Lupus erythematosus’ OR ‘Lupus Vulgaris’
#26	‘Ischemic heart disease’/exp *OR* ‘heart disease’/exp OR ‘Cardiovascular disease’/exp OR ‘Heart failure’/exp OR ‘Ischemic heart disease’ *OR* ‘heart disease’ OR ‘Cardiovascular disease’ OR ‘Heart failure’
#27	‘Crohn disease’/exp *OR* IBD *OR* ‘Irritable colon’/exp *OR* ‘Ulcerative colitis’/exp OR ‘Gastrointestinal Disease’/exp OR ‘Inflammatory bowel disease’/exp OR ‘Crohn disease’ *OR* ‘Irritable colon’ *OR* ‘Ulcerative colitis’ OR ‘Gastrointestinal Disease’ OR ‘Inflammatory bowel disease’
#28	‘Cystic fibrosis’/exp OR ‘Cystic fibrosis’
#29	‘Diabetes mellitus’/exp OR ‘Diabetes mellitus’ OR ‘Diabet’
#30	‘Dermatitis’/exp OR ‘Dermatitis’
#31	‘Endocrine disease’/exp OR ‘Endocrine disease’
#32	‘Endometriosis’/exp or ‘Endometriosis’
#33	Epilepsy/exp OR ‘seizure, epilepsy and convulsion’/exp OR Epilepsy OR ‘seizure, epilepsy and convulsion’
#34	Fibromyalgia/exp OR ‘Muscle disease’/exp OR Fibromyalgia OR ‘Muscle disease’
#35	‘Hemophilia’/exp OR haemophil* OR hemophil* or Hemophilia
#36	‘Hepatitis B’/exp OR ‘Hepatitis’/exp OR ‘Liver disease’/exp OR ‘Hepatitis B’ OR ‘Hepatitis’ OR ‘Liver disease’
#37	‘Human immunodeficiency virus infection’/exp *OR* ‘Human immunodeficiency virus’/exp OR ‘Human immunodeficiency virus infection’ *OR* ‘Human immunodeficiency virus’
#38	‘Hypertension’/exp *OR* ‘hypotension’/exp OR ‘Hypertension’ *OR* ‘hypotension’
#39	‘Lung disease’/exp *OR* ‘Lung fibrosis’/exp OR ‘Lung disease’ *OR* ‘Lung fibrosis’
#40	'Neurometabolic disorder' *OR* 'Metabolic disorder'/exp OR 'Metabolic disorder'
#41	‘Migraine’/exp OR ‘Headache’/exp OR ‘Muscle contraction headache’ OR Migraine OR Headache
#42	‘Neuralgia’/exp OR ‘Trigeminus neuralgia’/exp OR Neuralgia OR ‘Trigeminus neuralgia’
#43	‘Neuromuscular disease’/exp *OR* ‘Muscular dystrophy’/exp *OR* ‘Muscular dystrophy’ OR ‘Myasthenia Gravis’/exp OR 'neurologic disease'/exp OR ‘Muscle disease’/exp OR ‘Neuromuscular disease’ *OR* ‘Myasthenia Gravis’ OR 'neurologic disease' OR ‘Muscle disease’
#44	‘Otitis media'/exp OR ‘Otitis media'
#45	‘Peripheral occlusive artery disease’/exp OR ‘Peripheral vascular disease’/exp OR ‘Peripheral occlusive artery disease’ OR ‘Peripheral vascular disease’
#46	‘Phenylketonuria’/exp OR ‘Phenylketonuria’ OR PKU
#47	‘Ovary polycystic disease’/exp OR ‘Ovary polycystic disease’
#48	‘Psoriasis’/exp OR ‘Skin disease’/exp OR Psoriasis OR ‘Skin disease’
#49	‘Sickle cell anemia’/exp OR ‘Sickle cell anemia’
#50	‘Sjoegren syndrome’/exp OR ‘Sjoegren syndrome’
#51	‘Sleep disorder’/exp OR Insomnia/exp OR Narcolepsy/exp OR ‘Sleep disordered breathing’/exp OR ‘Sleep disorder’ OR Insomnia OR Narcolepsy OR ‘Sleep disordered breathing’
#52	‘Spinal cord disease’/exp *OR* ‘Spine disease’/exp *OR* ‘Spine injury’/exp *OR* ‘Spinal dysraphism’/exp *OR* ‘Spina bifida aperta’ *OR* ‘Spinal cord injury’/exp OR ‘Spinal cord disease’ *OR* ‘Spine disease’ *OR* ‘Spine injury’ *OR* ‘Spinal dysraphism’ *OR* ‘Spina bifida aperta’ OR ‘Spina bifida aperta’/exp *OR* ‘Spinal cord injury’
#53	‘Thalassemia’/exp OR Thalassemia
#54	‘Thyroid disease’/exp OR ‘Thyroid disease’
#55	Combine searches #4 - #54 using OR

#56	Combine searches #3 AND #55
#57	'Prevalence'/exp OR prevalen* OR 'prevalence'
#58	Combine searches #56 AND #57

Web of Science: Sources Medline

Filters:

Date Range: 2000 – 2021

Species: Humans

Age: All Child 0 – 18 years

Language: English

MESH Headings: Not Animals; Not Aged 80 and over; Not Middle aged

Refined by MeSH Headings: Fatigue

TS = Topic Search (includes title, abstract, keyword and index fields)

#1	TS=adolescen* OR TS=child* OR TS="young pe*" OR TS=teen* OR TS=juvenile* OR TS=youth* OR TS=minor* OR TS=puberty OR TS=pubescent* OR TS=prepubescent* OR TS=school* OR TS="high school" OR TS= "high-school" OR TS= "secondary school" OR TS="school child*" OR TS="schoolchild*" OR TS="school-child*" OR TS=paediatric* OR TS=pediatric* OR TS=boy* OR TS=girl* OR TS= "primary school" OR TS= "primary-school"
#2	TS=fatigue* OR TS= "lack of energy" OR TS= "loss of energy" OR TS= "low energy" OR TS=tired* OR TS=drows* OR TS=lethargy OR TS=lethargic OR TS=exhaust* OR TS=weary OR TS=weariness OR TS=drained OR TS=sleepy OR TS=sleepiness OR TS=lassitude OR TS=sluggish* OR TS=apathy OR TS=apathetic OR TS=astheni* OR TS=malaise
#3	#1 AND #2
#4	TS= "Chronic Illness" OR TS= "Chronic disease*" OR TS= "Chronic condition*" OR TS= "Chronic disorder*" OR TS="Chronic illness*" OR TS= "Chronic syndrome*" OR TS= "Chronic health" OR TS= "Long-term condition*" OR TS= "Long-term disease*" OR TS= "Long-term disorder*" OR TS= "Long-term illness*" OR TS= "Long-term syndrome*" OR TS= "Long term condition*" OR TS= "Long term disease*" OR TS= "Long term disorder*" OR TS= "Long term illness*" OR TS= "Long term syndrome*" OR TS= "Longstanding Condition*" OR TS= "Longstanding disease*" OR TS= "Longstanding illness*" OR TS= "Longstanding syndrome*" OR TS= "Longstanding disorder*" OR TS= "Long-standing condition*" OR TS= "Long-standing disease*" OR TS= "Long-standing disorder*" OR TS= "Long-standing illness*" OR TS= "Long-standing syndrome*"
#5	TS= AIDS OR TS= "Acquired immunodeficiency syndrome"
#6	TS= "Allergic Rhinitis" OR TS= "Hay fever" OR TS= Hay-fever OR TS="Respiratory Tract Disorders"
#7	TS= "Allergic disorders" OR TS= Allerg* OR TS= "Allergic Skin Disorders"
#8	TS= Anemia OR TS= "Blood and Lymphatic Disorders" OR TS= "Genetic Disorders" OR TS= Anaem* OR TS= "Fanconi Anaemia" OR TS= Anem* OR TS= "Fanconi Anemia"

#9	TS= Angina OR TS="Heart Disorders"
#10	TS= Arrhythm*
#11	TS= "Rheumatoid Arthritis" OR TS= Arthriti* OR TS= "Juvenile Arthritis"
#12	TS=Asthma*
#13	TS= "Atrial fibrillation"
#14	TS= "Immunologic Disorder*" OR TS= "Autoimmune Disease*"
#15	TS= Neoplasms OR TS= Cancer* OR TS= Oncolog*
#16	TS= "Cerebral Palsy" OR "Brain Disorder*" OR "Central Nervous System Disorder*"
#17	TS= Gout* OR TS= "Gouty Arthritis"
#18	TS= "Kidney disease*" OR TS= "Kidney failure"
#19	TS="Chronic Obstructive Pulmonary Disease" OR TS= "COPD" OR TS= "Chronic Pulmonary Disease" OR TS= "Lung Disorder*"
#20	TS= "Chronic pain"
#21	TS= Sarcoidosis OR TS= sarcoid*
#22	TS= "Congenital Disorder*" OR TS= "Neonatal Disorder*" OR TS= "Cleft Lip" OR TS= "Cleft Palate"
#23	TS= Coeliac* OR TS= Celiac*
#24	TS= "Connective tissue disease*" OR TS= Lupus
#25	TS="Heart Disorder*" OR TS= "Cardiovascular Disorder*" OR TS= "Coronary Heart Disease*" OR TS= "Heart Disease*"
#26	TS= "Colon Disorder*" OR TS= "Gastrointestinal disorder*" OR TS= "crohn's" OR TS= "Ulcerative colitis" OR TS= "inflammatory bowel disease*" OR TS= "IBD"
#27	TS= "Cystic Fibrosis"
#28	TS= Diabet*
#29	TS= Dermatitis OR TS= Eczema*
#30	TS= "Endocrine Disease*" OR TS= Thyroid OR TS= Adrenal OR TS="Endocrine disorder*"
#31	TS= Endometri*
#32	TS= Epilep* OR TS= Seizure*
#33	TS= "Muscular Disorder*" OR TS= Fibromyalgia
#34	TS= Haemophil* OR TS= Hemophil*
#35	TS= "Heart failure" OR TS= "Heart condition*" OR TS= "Heart Disease*" OR TS= "Heart Defect*"
#36	TS= Hepatitis OR TS="Liver Disorder*"
#37	TS= "HIV" OR TS= "Human Immunodeficiency Virus"

#38	TS= Hypertens* OR TS= Hypotens* OR TS= “High Blood Pressure” OR TS= “Blood pressure disorder*”
#39	TS= “Lung disease*” OR TS= “Lung disorder*” OR TS= “Lung fibrosis”
#40	TS= “Neurometabolic disorder*” OR TS= “Neurometabolic condition*” OR TS= “Neurometabolic disease*” OR TS= Metabolic OR TS= “Metabolic Syndrome”
#41	TS= Migraine OR TS=Headache
#42	TS= Neuralg*
#43	TS= “Nervous System Disorders” OR TS= “Muscular Dystrophy” OR TS= “Myasthenia Gravis” OR TS= “Muscular Disorder*” OR TS= “Musculoskeletal Disorder*” OR TS= “Neuromuscular Disease*” OR TS= “Neuromuscular Condition*” OR TS= “Neuromuscular Disorder*”
#44	TS= "Chronic otitis media" OR TS= “Otitis media”
#45	TS= "Peripheral Arterial Disease" OR TS= "peripheral vascular disease"
#46	TS= Phenylketonuria OR TS= “PKU” OR “Metabolism Disorder*” OR “Metabolic Disorder*”
#47	TS= "Polycystic ovary syndrome" OR TS= “polycystic ovaries” OR TS= “PCOS”
#48	TS= Psoriasis OR TS= “Skin Disorder*”
#49	TS= “Sickle cell disease*”
#50	TS= Sjörgen* OR TS= Sjorgen
#51	TS= “Sleep disorder*” OR TS= “Sleep apnoea” OR TS= Narcolepsy OR TS= “Sleep apnea” OR TS= “Sleep wake disorder*” OR TS= Insomnia
#52	TS= “Spina bifida”
#53	TS= “Spinal cord injur*” OR TS= “Spinal disease*” OR TS= “Spinal injur*”
#54	TS= Thalassem*
#55	TS= Hyperthyroid* OR TS= Hypothyroid* OR TS= “Thyroid disorder*“
#56	Combine searches 4 – 55 using OR
#57	Combine searches 3 AND 56
#58	TS= prevalence OR TS= prevalen* OR TS= risk* OR TS= rate* OR TS= trend OR TS= associat*
#59	Combine searches #58 AND #57

Appendix C

Literature Review Adapted Quality Assessment Tool

Question	Answer	Points
Selection: 1. Representativeness of the sample	a. Population contained a mixture of specialities or recruited through multiple sites	1
	b. Population contained a single chronic health condition at a single site	0
Selection: 2. Sample Size	a. Sample size is justified and satisfactory	1
	b. No information provided or not justified	0
Selection: 3. Non-respondents	a. Comparability between respondents and non-respondents characteristics is established, and the response rate is satisfactory	1
	b. Response rate is unsatisfactory, or the comparability between respondents and non-respondents is unsatisfactory, or there was no description of the response rate or the characteristics of responders and the non-responders	0
Selection: 4. Ascertainment of the exposure (risk factor) i.e. the chronic health condition	a. Clinic registers or hospital records or validated measurement tool e.g. MRI/Medical tests **	1
	b. Parental/Personal report	0
	c. No description/No information	0
Outcome: 5. Assessment of outcome (fatigue prevalence)	a. Validated measure of fatigue, using a validated cut off score	1
	b. Non-validated measurement tool, or validated measurement tool with non-valid cut off score	0
Outcome: 6. Quality of descriptive statistics reporting	a. Reported descriptive statistics to describe the population (e.g. age, sex) with proper measures of dispersion (e.g. standard deviation, standard error, range, confidence intervals) e.g., reported mean scores of fatigue with range/error/CI	1
	b. Descriptive statistics were not reported, were incomplete, or did not include proper measures of dispersion	0

Service-Related Project Appendices

Appendix D

Service-Related Project Author Guidelines - Mindfulness Journal

Instructions for Authors

Double-blind peer review

This journal follows a double-blind reviewing procedure. This means that the author will remain anonymous to the reviewers throughout peer review. It is the responsibility of the author to anonymize the manuscript and any associated materials.

Author names, affiliations and any other potentially identifying information should be removed from the manuscript text and any accompanying files (such as figures of supplementary material);

A separate Title Page should be submitted, containing title, author names, affiliations, and the contact information of the corresponding author. Any acknowledgements, disclosures, or funding information should also be included on this page;

Authors should avoid citing their own work in a way that could reveal their identity.

Manuscript Submission

Manuscript Submission

Submission of a manuscript implies: that the work described has not been published before; that it is not under consideration for publication anywhere else; that its publication has been approved by all co-authors, if any, as well as by the responsible authorities – tacitly or explicitly – at the institute where the work has been carried out. The publisher will not be held legally responsible should there be any claims for compensation.

Permissions

Authors wishing to include figures, tables, or text passages that have already been published elsewhere are required to obtain permission from the copyright owner(s) for both the print and online format and to include evidence that such permission has been granted when submitting their papers. Any material received without such evidence will be assumed to originate from the authors.

Online Submission

Please follow the hyperlink “Submit manuscript” and upload all of your manuscript files following the instructions given on the screen.

Source Files

Please ensure you provide all relevant editable source files at every submission and revision. Failing to submit a complete set of editable source files will result in your article

not being considered for review. For your manuscript text please always submit in common word processing formats such as .docx or LaTeX.

Suggested Reviewers

Authors of research and review papers, excluding editorial and book review submissions, are allowed to provide the names and contact information for, maximum, 4 to 6 possible reviewers of their paper. When uploading a paper to the Editorial Manager site, authors must provide complete contact information for each recommended reviewer, along with a specific reason for your suggestion in the comments box for each person. The journal will consider reviewers recommended by the authors only if the reviewers' institutional email is provided. A minimum of two suggested reviewers should be from a university or research institute in the United States. You may not suggest the Editor or Associate Editors of the journal as potential reviewers. Although there is no guarantee that the editorial office will use your suggested reviewers, your help is appreciated and may speed up the selection of appropriate reviewers.

Authors should note that it is inappropriate to list as preferred reviewers researchers from the same institution as any of the authors, collaborators and co-authors from the past five years as well as anyone whose relationship with one of the authors may present a conflict of interest. The journal will not tolerate this practice and reserves the right to reject submissions on this basis.

Title Page

The title page should include:

The name(s) of the author(s)

A concise and informative title

The affiliation(s) and address(es) of the author(s)

The e-mail address, and telephone number(s) of the corresponding author

If available, the 16-digit ORCID of the author(s)

Abstract

Please provide of structured abstract of up to 250 words

Keywords

Please provide 4 to 6 keywords which can be used for indexing purposes.

Structured Abstract

The structured abstract of up to 250 words with four labeled sections should containing the following, with sub-section headers in bold:

a. Objectives: Problem being addressed in the study

b. Methods: The participants, essential features of the study method

c. Results: The basic findings, including effect sizes and confidence intervals and/or statistical significance levels

d. Conclusions: What the authors conclude from study results

Text

Text Formatting

Manuscripts should be submitted in Word.

Use a normal, plain font (e.g., 12-point Times Roman) for text.

Use italics for emphasis.

Use the automatic page numbering function to number the pages.

Do not use field functions.

Use tab stops or other commands for indents, not the space bar.

Use the table function, not spreadsheets, to make tables.

Use the equation editor or MathType for equations.

Save your file in docx format (Word 2007 or higher) or doc format (older Word versions).

Headings

Please use no more than three levels of displayed headings.

Abbreviations

Abbreviations should be defined at first mention and used consistently thereafter.

Acknowledgments

Acknowledgments of people, grants, funds, etc. should be placed in a separate section on the title page. The names of funding organizations should be written in full.

Footnotes

This journal does not allow the use of footnotes, except in reprinted papers.

Article length

Papers accepted for publication in this journal are 45 double-spaced pages, in 12-point font, inclusive of text, references, tables and figures. For manuscripts exceeding this length, authors should contact the Editor in Chief, Nirbhay N. Singh directly at nirbz52@gmail.com.

Terminology

• Please always use internationally accepted signs and symbols for units (SI units).

Scientific style

Generic names of drugs and pesticides are preferred; if trade names are used, the generic name should be given at first mention.

Please use the standard mathematical notation for formulae, symbols etc.:Italic for single letters that denote mathematical constants, variables, and unknown quantities Roman/upright for numerals, operators, and punctuation, and commonly defined functions or abbreviations, e.g., cos, det, e or exp, lim, log, max, min, sin, tan, d (for derivative) Bold for vectors, tensors, and matrices.

References

Citation

Cite references in the text by name and year in parentheses. Some examples:

Negotiation research spans many disciplines (Thompson, 1990).

This result was later contradicted by Becker and Seligman (1996).

This effect has been widely studied (Abbott, 1991; Barakat et al., 1995; Kelso & Smith, 1998; Medvec et al., 1999).

Authors are encouraged to follow official APA version 7 guidelines on the number of authors included in reference list entries (i.e., include all authors up to 20; for larger groups, give the first 19 names followed by an ellipsis and the final author's name). However, if authors shorten the author group by using et al., this will be retained.

Reference list

The list of references should only include works that are cited in the text and that have been published or accepted for publication. Personal communications and unpublished works should only be mentioned in the text.

Reference list entries should be alphabetized by the last names of the first author of each work.

Journal names and book titles should be *italicized*.

If available, please always include DOIs as full DOI links in your reference list (e.g. "https://doi.org/abc").

Journal article Grady, J. S., Her, M., Moreno, G., Perez, C., & Yelinek, J. (2019). Emotions in storybooks: A comparison of storybooks that represent ethnic and racial groups in the United States. *Psychology of Popular Media Culture, 8*(3), 207–217. https://doi.org/10.1037/ppm0000185

Article by DOI Hong, I., Knox, S., Pryor, L., Mroz, T. M., Graham, J., Shields, M. F., & Reistetter, T. A. (2020). Is referral to home health rehabilitation following inpatient rehabilitation facility associated with 90-day hospital readmission for adult patients with stroke? *American Journal of Physical Medicine & Rehabilitation*. Advance online publication. https://doi.org/10.1097/PHM.0000000000001435

Book Sapolsky, R. M. (2017). *Behave: The biology of humans at our best and worst*. Penguin Books.

Book chapter Dillard, J. P. (2020). Currents in the study of persuasion. In M. B. Oliver, A. A. Raney, & J. Bryant (Eds.), *Media effects: Advances in theory and research* (4th ed., pp. 115–129). Routledge.

Online document Fagan, J. (2019, March 25). *Nursing clinical brain*. OER Commons. Retrieved January 7, 2020, from https://www.oercommons.org/authoring/53029-nursing-clinical-brain/view

Please note:

If you are citing journal articles by their DOI please make sure to also include the volume and page numbers, if already available, e. g. as follows: “Slifka, M. K., & Whitton, J. L. (2000) Clinical implications of dysregulated cytokine production. Journal of Molecular Medicine, 78(2), 74-80. https://doi.org/10.1007/s001090000086”.

Tables

All tables are to be numbered using Arabic numerals.

Tables should always be cited in text in consecutive numerical order.

For each table, please supply a table caption (title) explaining the components of the table.

Identify any previously published material by giving the original source in the form of a reference at the end of the table caption.

Footnotes to tables should be indicated by superscript lower-case letters (or asterisks for significance values and other statistical data) and included beneath the table body.

Artwork and Illustrations Guidelines

Electronic Figure Submission

Supply all figures electronically.

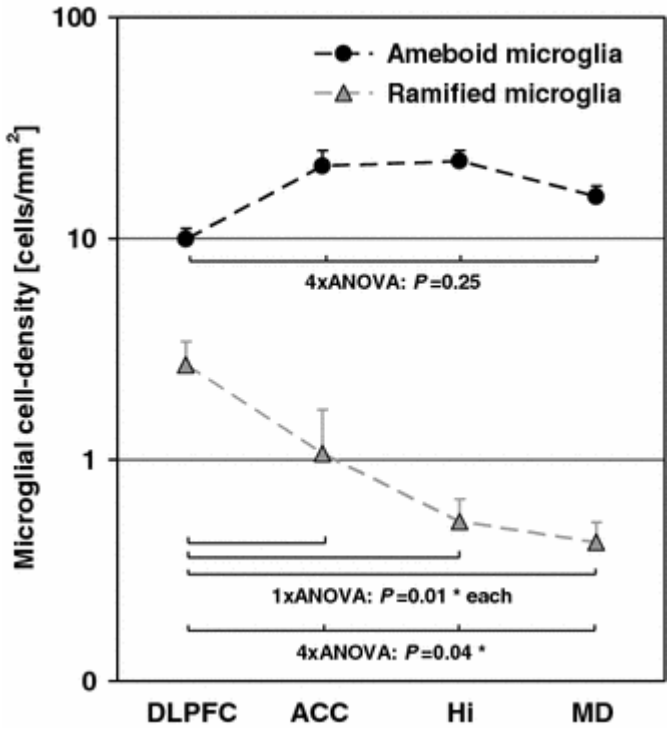
Indicate what graphics program was used to create the artwork.

For vector graphics, the preferred format is EPS; for halftones, please use TIFF format. MSOffice files are also acceptable.

Vector graphics containing fonts must have the fonts embedded in the files.

Name your figure files with "Fig" and the figure number, e.g., Fig1.eps.

Line Art



Definition: Black and white graphic with no shading.

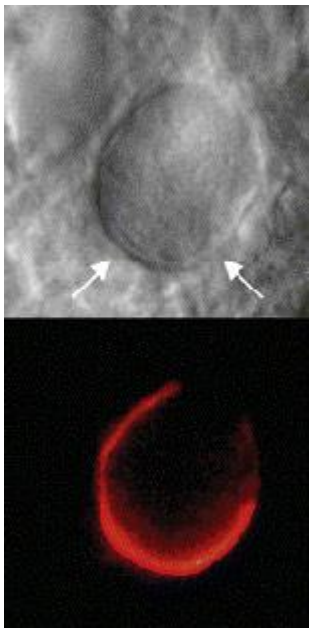
Do not use faint lines and/or lettering and check that all lines and lettering within the figures are legible at final size.

All lines should be at least 0.1 mm (0.3 pt) wide.

Scanned line drawings and line drawings in bitmap format should have a minimum resolution of 1200 dpi.

Vector graphics containing fonts must have the fonts embedded in the files.

Halftone Art

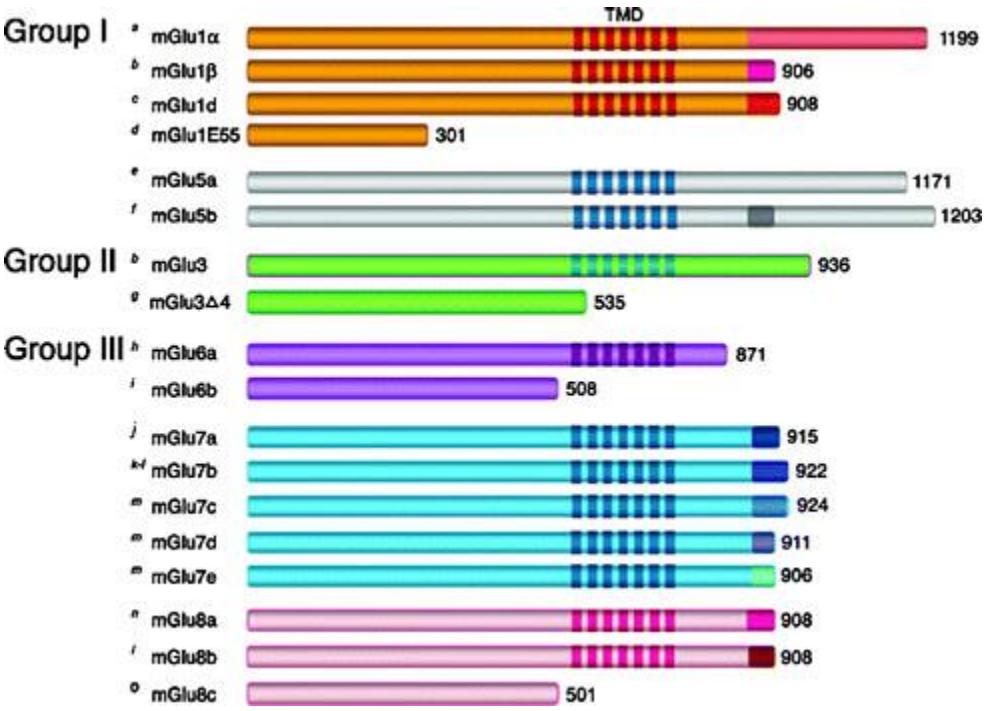


Definition: Photographs, drawings, or paintings with fine shading, etc.

If any magnification is used in the photographs, indicate this by using scale bars within the figures themselves.

Halftones should have a minimum resolution of 300 dpi.

Combination Art



Definition: a combination of halftone and line art, e.g., halftones containing line drawing, extensive lettering, color diagrams, etc.

Combination artwork should have a minimum resolution of 600 dpi.

Color Art

Color art is free of charge for online publication.

If black and white will be shown in the print version, make sure that the main information will still be visible. Many colors are not distinguishable from one another when converted to black and white. A simple way to check this is to make a xerographic copy to see if the necessary distinctions between the different colors are still apparent.

If the figures will be printed in black and white, do not refer to color in the captions.

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Figure Lettering

To add lettering, it is best to use Helvetica or Arial (sans serif fonts).

Keep lettering consistently sized throughout your final-sized artwork, usually about 2–3 mm (8–12 pt).

Variance of type size within an illustration should be minimal, e.g., do not use 8-pt type on an axis and 20-pt type for the axis label.

Avoid effects such as shading, outline letters, etc.

Do not include titles or captions within your illustrations.

Figure Numbering

All figures are to be numbered using Arabic numerals.

Figures should always be cited in text in consecutive numerical order.

Figure parts should be denoted by lowercase letters (a, b, c, etc.).

If an appendix appears in your article and it contains one or more figures, continue the consecutive numbering of the main text. Do not number the appendix figures,"A1, A2, A3, etc." Figures in online appendices [Supplementary Information (SI)] should, however, be numbered separately.

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Each figure should have a concise caption describing accurately what the figure depicts. Include the captions in the text file of the manuscript, not in the figure file.

Figure captions begin with the term Fig. in bold type, followed by the figure number, also in bold type.

No punctuation is to be included after the number, nor is any punctuation to be placed at the end of the caption.

Identify all elements found in the figure in the figure caption; and use boxes, circles, etc., as coordinate points in graphs.

Identify previously published material by giving the original source in the form of a reference citation at the end of the figure caption.

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Figures should be submitted separately from the text, if possible.

When preparing your figures, size figures to fit in the column width.

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In order to give people of all abilities and disabilities access to the content of your figures, please make sure that

All figures have descriptive captions (blind users could then use a text-to-speech software or a text-to-Braille hardware)

Patterns are used instead of or in addition to colors for conveying information (colorblind users would then be able to distinguish the visual elements)

Any figure lettering has a contrast ratio of at least 4.5:1

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Springer accepts electronic multimedia files (animations, movies, audio, etc.) and other supplementary files to be published online along with an article or a book chapter. This feature can add dimension to the author's article, as certain information cannot be printed or is more convenient in electronic form.

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High resolution (streamable quality) videos can be submitted up to a maximum of 25GB; low resolution videos should not be larger than 5GB.

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Maximum file size: 25 GB for high resolution files; 5 GB for low resolution files

Minimum video duration: 1 sec

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It is possible to collect multiple files in a .zip or .gz file.

Numbering

If supplying any supplementary material, the text must make specific mention of the material as a citation, similar to that of figures and tables.

Refer to the supplementary files as “Online Resource”, e.g., "... as shown in the animation (Online Resource 3)", “... additional data are given in Online Resource 4”.

Name the files consecutively, e.g. “ESM_3.mpg”, “ESM_4.pdf”.

Captions

For each supplementary material, please supply a concise caption describing the content of the file.

Processing of supplementary files

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Accessibility

In order to give people of all abilities and disabilities access to the content of your supplementary files, please make sure that

The manuscript contains a descriptive caption for each supplementary material

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Ethical standards

Manuscripts submitted for publication must contain a statement to the effect that all human and animal studies have been approved by the appropriate ethics committee and have therefore been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments.

It should also be stated clearly in the text that all persons gave their informed consent prior to their inclusion in the study. Details that might disclose the identity of the subjects under study should be omitted.

These statements should be added in a separate section before the reference list. If these statements are not applicable, authors should state: The manuscript does not contain clinical studies or patient data.

The editors reserve the right to reject manuscripts that do not comply with the above-mentioned requirements. The author will be held responsible for false statements or failure to fulfill the above-mentioned requirements

Conflict of interest

Authors must indicate whether or not they have a financial relationship with the organization that sponsored the research. This note should be added in a separate section before the reference list.

If no conflict exists, authors should state: The authors declare that they have no conflict of interest.

English Language Editing

For editors and reviewers to accurately assess the work presented in your manuscript you need to ensure the English language is of sufficient quality to be understood. If you need help with writing in English you should consider:

Getting a fast, free online grammar check.

Asking a colleague who is proficient in English to review your manuscript for clarity.

Visiting the English language tutorial which covers the common mistakes when writing in English.

Using a professional language editing service where editors will improve the English to ensure that your meaning is clear and identify problems that require your review. Two such services are provided by our affiliates Nature Research Editing Service and American Journal Experts.

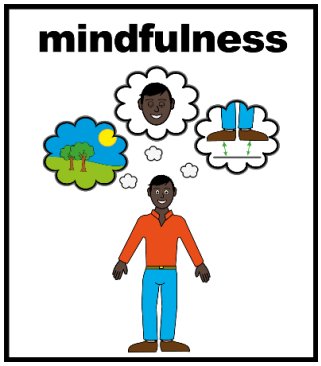
Please note that the use of a language editing service is not a requirement for publication in this journal and does not imply or guarantee that the article will be selected for peer review or accepted.

If your manuscript is accepted it will be checked by our copyeditors for spelling and formal style before publication.

Appendix E

Service-Related Project Plain English Summary

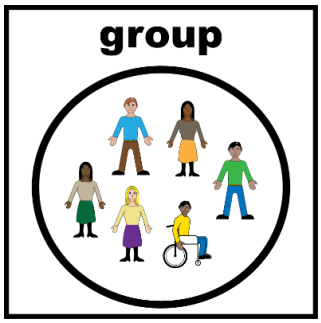
Background



‘Mindful Life’ is a mindfulness group for adults, aged 18 and above, who experience mental health difficulties.

Mindfulness means paying attention to the present moment and being aware of your thoughts, feelings, and the world around you.

Previous research shows that learning mindfulness-based skills can improve mood and wellbeing for those with and without mental health difficulties.



The Mindful Life group is run by an NHS mental health service for those with a range of difficulties, such as anxiety, depression, psychosis, or Alzheimer’s Disease.

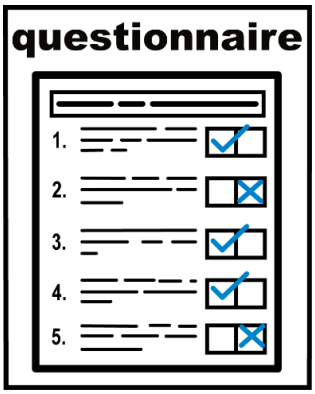
The group involves 12 sessions which take place once a week, for 12 weeks. It aims to help people develop mindfulness-based skills and reduce feelings of distress.

In 2019, a researcher interviewed 8 people who had previously attended the Mindful Life group.

Most said that the group helped to learn how to manage distress. They also said groups were supportive and the facilitators were understanding.

Research Aims

This research aimed to build on the interviews by exploring whether mindfulness-based skills and distress levels changed over the course of the group.



Those who attended the group were asked to complete two questionnaires at the start and again at the end of the group.

One questionnaire measured levels of distress.

The other questionnaire measured mindfulness-based skills. This gave us a total mindfulness score and scores for five specific mindfulness-based skills.

Based on these questionnaires, the research aimed to answer four questions:

- For those who attend the group, do levels of distress reduce from the beginning to the end of the group?
- Do mindfulness-based skills increase from the beginning to end of the group?
- Is there a relationship between total mindfulness scores and distress levels?
- Are there any specific areas of mindfulness that are linked with reduced levels of distress?

What did we find?

- Between 2015 and 2020, 201 people were referred to the group and invited to meet the facilitators for an assessment.
- 111 people were invited to start the group and 70 people completed the group.
- 50 people completed the questionnaires at the start and end of the group, although not everyone completed both questionnaires.

Did levels of distress reduce from the beginning to the end of the group?

- Based on average scores, there was a slight decrease in levels of distress from start to end of the group, however it was not a significant change.

- This means that we cannot confidently say that the group helps to reduce levels of distress for everyone.

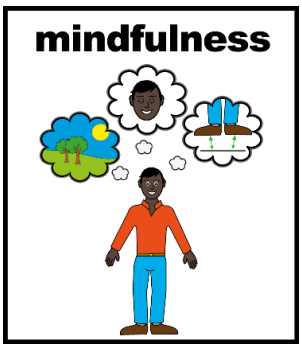
Why might that be?

- We do not know whether stressful life events or personal circumstances affected distress levels over the 12 weeks.

- Mindfulness encourages people to pay attention to thoughts and feelings so some people may become more aware of their difficulties.

- Practicing mindfulness between sessions is key but we do not know how many people did this.

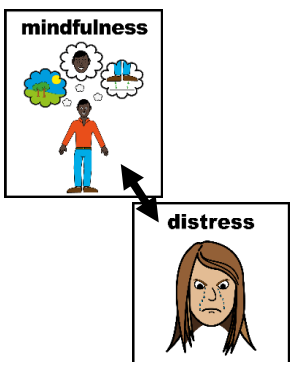
Did mindfulness-based skills increase?



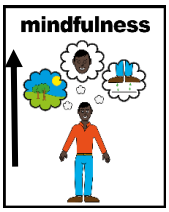
- Total mindfulness scores increased as did scores in three specific mindfulness-based skills.

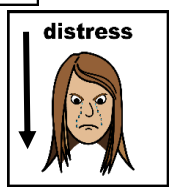
- Two other areas of mindfulness were collected within the questionnaire and did not change significantly over the 12 weeks.

Is there a relationship between total mindfulness scores and distress levels?



- There was a small suggestion that as overall mindfulness-based skills increase, levels of distress reduce. This was not significant which means we cannot be confident that there is a link between them.





Are there any specific mindfulness-based skills that are associated with reduced levels of distress?

- One specific mindfulness-based skill focuses on not reacting to thoughts, feelings or internal sensations, i.e., noticing them but not trying to change or get rid of them. We found that as this skill increased, ratings of distress appeared to decrease.

- We cannot be sure that one causes the other, but this finding is in line with previous research which has looked at how mindfulness-based interventions work.

What can the service do next?

- **Work with other teams.** We found that the Early Intervention for Psychosis team referred the fewest number of people to the group. This may mean that some people are missing out on an intervention that they might find helpful.

- **Track next steps after attending the group.** To understand more about the impact of the group and benefits for the service and service users, it could be helpful to see if people who attend are discharged or access other interventions.

- **Increase key practices.** Helping people to develop skills in ‘not reacting to thoughts or feelings’ may help to further reduce levels of distress.

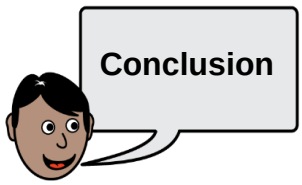
- **Support people to complete questionnaires, at start and end of the group.** Offering options to support people to complete questionnaires e.g., in session, at home, or with phone support from facilitator, will help the service to continue evaluating and improving the group.

- **Look at why people stop attending the group.** Understanding why some people choose not to attend or stop attending the group could help to problem solve any difficulties or barriers and help make the group more accessible for people.

- **Collect information on mindful practices at home.** We do not know how many people practice mindfulness-based skills between group sessions, or whether this makes a difference to levels of distress. Collecting this information could help to encourage and tailor home-based practices and may help to reduce distress.

- **Compare face-to-face with online groups.** The information for this project was recorded before the COVID-19 pandemic and the groups took place in person. The group now takes place online. Evaluating the differences will be helpful to decide what works best for service users.

Conclusions



- Mindfulness-based skills increased over the course of the 12-week group and there was a small but not-significant decrease in levels of distress over this time.

- A key mindfulness-based skill which appears to be linked with reduced distress is not reacting to internal experiences. Practices that focus on building this mindfulness-based skill may therefore help people to experience less distress which will be important for facilitators and service users to be aware of when joining the group.

Appendix F

Service-Related Project Ethical Approval

NHS
Avon and Wiltshire
Mental Health Partnership
NHS Trust

Kiesha Williams

University of Bath

Carla Carter
Quality Improvement &
Clinical Audit Manager
AWP NHS Trust
Victoria Centre
53 Downs Way
Swindon SN3 6BW

T: 01793 327876
Or dial reception on:
01793 327800

Date: May 2020

Dear Kiesha

Re: Evaluation of a Mindful Life Group

I am pleased to confirm approval of your Service Evaluation by AWP NHS Trust.

Please note that this approval has come from AWP's Quality Team and not AWP's Research and Development Team. However, we do expect a good level of governance will be achieved from the ethical scrutiny by your University as well as adherence to general ethical principles for the protection of patients. The specific ethical principles and patient protection laws to be followed are:

- **Consent** – It is important that potential participants are not coerced to take part in the project. They have the right to refuse to take part and to withdraw at any point and this is explained via an information sheet provided prior to any engagement or data gathering such as surveys or interviews. This information sheet will often lead to the signing of a consent form by participants agreeing to take part in your Project.

- **Anonymity** – Participants need to know whether their anonymity will be protected and if so how this will be carried out. This will also be documented within your participants' information sheet/consent form.

- **Data protection and privacy** – You need to consider how you are going to ensure that your data is stored safely and that participant privacy is protected. Again this should be stipulated within your participants' information sheet/consent form. You will need to adhere to the Data Protection Act (2018) and the General Data Protection Regulation (GDPR).

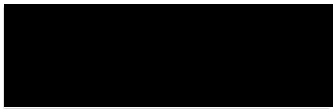
Jess Hillier (jess.hillier@nhs.net) has been assigned as your allocated AWP Quality Team Facilitator. Please contact this Facilitator if you have any queries or require further support or information during your project. They will email you at regular intervals for updates, so that progress of your project can be updated on our central project database, and fed into Trust

committees. You will be assigned an AWP Project Reference Number by your Facilitator once we have confirmation that data collection has commenced and the project has actually started.

The importance of dissemination of all Service Evaluation or Quality Improvement work cannot be over emphasised. For this reason, the findings of all Projects should be shared with the Quality Team so that we can make judgements regarding risk and champion and disseminate the results across the rest of the Trust so that good practice can be shared and replication kept to a minimum. Reports may require approval by Locality Governance Groups if specific actions or improvements are required following your findings or particularly, if you wish to gain external publication, you will require AWP approval of your final report before doing so. Therefore, please share draft copies of your report with your Facilitator so that presentation and approval at Governance Groups can be arranged. Once you have an approved final version of your report, please ensure you send a copy to your allocated Facilitator.

If you do need any further support or information, please contact your Facilitator or myself, quoting the title of your project.

Yours sincerely



Carla Carter
Quality Improvement and Clinical Audit Manager

Appendix G

Main Research Project Author Guidelines - Journal of Neuromuscular Disorders

Author Information

Types of Paper

Research Articles
Regular original research articles should be sent to the main Editorial Office. There is no restriction on length though most articles are between 2500 and 6000 words long. Please contact the Editorial Office if you wish to discuss. The Editor-in-Chief or an appropriate Executive Associate Editor will handle the submission. (For more information on Executive Associate Editors please see Editor's Commentary. Neuromuscular Disorders, Volume 26, Issue 1, January 2016, Pages 1–4.)

Animal Models for Neuromuscular Diseases
Gillian Butler-Browne will be allocated research articles submitted under this section. There is no restriction on length though most articles are between 2500 and 6000 words long. Please contact the Editorial Office if you would like to discuss.

Veterinary Myology
Diane Shelton will be pleased to receive research articles covering clinical or investigative aspects of spontaneously occurring myopathies, neuropathies or disorders of neuromuscular transmission in domestic animals. There is no restriction on length though most articles are between 2500 and 6000 words long. Please contact the Editorial Office if you would like to discuss.

In addition to submitting regular original research articles, letters and meeting reports, we invite readers to submit interesting articles to the special sections listed below. All items should be submitted online in the usual way to the main Editorial Office in London, with the relevant article type selected from the drop-down menu. If you wish to discuss anything with section editors prior to submission please refer to the journal homepage online or the inside front cover of the printed journal for up-to-date contact information of each section editor.

Reviews
Review papers should cover recent, important developments related to diagnosis, pathogenesis or therapy of a neuromuscular disorder. They can be either in-depth and comprehensive, or short, mini-reviews. Please include an abstract and key words. Reviews will be directed to Anders Oldfors who will co-ordinate peer review. There is no upper limit on the length though most articles do not exceed 6000 words. Please contact the Editorial Office if you would like to discuss.

Case Reports
Case reports should be of interest to the multidisciplinary readership of Neuromuscular Disorders and have a neuromuscular component. Topics such as sensory neuropathies and ataxias are of limited interest to our readership. Case reports should not exceed 2000

words and may include up to three tables or figures and a maximum of 25 references. They should take the form of Title, Abstract (up to 150 words), Introduction, Case Report, Discussion, Acknowledgements and References. Please note that key clinical information must be included in the abstract. Case reports will be directed to Beril Talim who will co-ordinate the editorial process.

Picture of the Month
Please send an interesting picture, clinical, pathological or imaging, of clinical challenge or interest. This should be accompanied by a brief case presentation and discussion, highlighting the special features of the picture, in no more than 300 words and up to three references (no abstract is required). The picture should be the main part of the presentation and be of adequate size and good quality.

Clinical Casebook
Contributions will be welcome for this section for cases that show a conflict of interpretation between the clinical and the investigative aspects of a case, with a view to raising questions, promoting thinking and discussion and potentially opening new channels of research to advance our knowledge.

Historical Reports
We welcome articles of historical interest. These can be sent to the Editorial Office in the first instance and will be redirected to the Historical Section Editor.

ENMC Workshop Reports
These submissions will be treated as a report on a workshop, with the convenor(s) listed as corresponding author(s). They will not be subjected to peer review and, after approval by the Editor, will be published in the next available issue of the journal. The workshop report should be concise and follow the agenda of the workshop - it has the nature of a workshop report, not of a review article (setting the stage for future developments).

The length of a report will vary depending on the number of topics discussed. Workshop reports need to be succinct, focusing on the new information. The references should be confined to those directly relevant to the workshop. Up to three tables, figures or photos may be included. No abstract is required.

1. The basic format of the ENMC-based workshop reports will be the same as in the past with a TITLE reflecting the number of the ENMC workshop, the number if appropriate of the topic workshop and the location and date.

2. A full list of all PARTICIPANTS will be included at the end of the report, with their city and country. This list will also include any ENMC representative as appropriate with [ENMC] after their name.

3. Full ACKNOWLEDGEMENT will be given to ENMC and all its sponsoring organisations at the end of the report using the exact wording as requested by ENMC as one of the conditions in their original letter of acceptance of the workshop.

4. In principle, only the workshop organizers will be the author(s) of the workshop report.

The organizers are to make sure that the tasks of all workshop participants regarding the preparation of the meeting report will have been discussed prior to closing the workshop.

All workshop participants will be included in the "ENMC XXXX Workshop Study Group*", so that they can be found in PubMed as co-authors of the workshop report. The workshop participants/report authors will be mentioned in an Appendix under the asterisk. The maximum number of authors for a workshop report (including the "ENMC study group") will be five – so a maximum of four (organizer) names can be used for the workshop report.

The list of authors will be included on the first page of the report, under the title, with a similar format to original papers in the journal. A full but preferably brief address can be included for each author, and the corresponding author for proofs and reprints should also be indicated.

5. As in the past, these reports will not be subjected to any peer review and it will be assumed that the content has the approval of all participants of the workshop. Once approved by the editor, the report will be given priority publication in the next available issue of the journal.

6. Keywords can be provided for reference.

Contact details for submission

Authors may send queries concerning the submission process, manuscript status or journal procedures to the Editorial Office (jane.miller@ucl.ac.uk).

Before you begin
Ethics in publishing

Please see our information on Ethics in publishing.

Description of variants (mutations)

Authors are required to follow the recommendations of the HGVS to describe sequence variants (see http://www.HGVS.org/mutnomen/ for a summary of the current recommendations).

Submission of data to a genetic database

In keeping with the recommendations of the Human Variome Project (Cotton RG et al 207. Nat Genet 39:433 http://www.nature.com/ng/journal/v39/n4/full/ng2024.html) authors submitting a manuscript to *Neuromuscular Disorders* are required to submit all variants and phenotype descriptions to a public database prior to acceptance. Authors must declare the status of database submission in their covering letter upon submission to the journal. In addition, authors should indicate in their manuscript the database(s) to which they have submitted the variants, and provide the URL. For further information and links to

gene variant databases either use GeneSymbol.lovd.nl (e.g. TP53.lovd.nl) or visit the following website: http://www.hgvs.org/dblist/dblist.html.

Informed consent and patient details

Studies on patients or volunteers require ethics committee approval and informed consent, which should be documented in the paper. Appropriate consents, permissions and releases must be obtained where an author wishes to include case details or other personal information or images of patients and any other individuals in an Elsevier publication. Written consents must be retained by the author but copies should not be provided to the journal. Only if specifically requested by the journal in exceptional circumstances (for example if a legal issue arises) the author must provide copies of the consents or evidence that such consents have been obtained. For more information, please review the Elsevier Policy on the Use of Images or Personal Information of Patients or other Individuals. Unless you have written permission from the patient (or, where applicable, the next of kin), the personal details of any patient included in any part of the article and in any supplementary materials (including all illustrations and videos) must be removed before submission.

Declaration of interest

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

Submission declaration and verification

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

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Contributors

Each author is required to declare his or her individual contribution to the article: all authors must have materially participated in the research and/or article preparation, so roles for all authors should be described. The statement that all authors have approved the final article should be true and included in the disclosure.

Changes to authorship

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

PDF file used in the peer-review process. Editable files (e.g., Word, LaTeX) are required to typeset your article for final publication. All correspondence, including notification of the Editor's decision and requests for revision, is sent by e-mail.

Submit your article

Please submit your article via Editorial Manager

Preparation
Queries

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

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

Use of word processing software

It is important that the file be saved in the native format of the word processor used. The text should be in single-column format. Keep the layout of the text as simple as possible. Most formatting codes will be removed and replaced on processing the article. In particular, do not use the word processor's options to justify text or to hyphenate words. However, do use bold face, italics, subscripts, superscripts etc. When preparing tables, if you are using a table grid, use only one grid for each individual table and not a grid for each row. If no grid is used, use tabs, not spaces, to align columns. The electronic text should be prepared in a way very similar to that of conventional manuscripts (see also the Guide to Publishing with Elsevier). Note that source files of figures, tables and text graphics will be required whether or not you embed your figures in the text. See also the section on Electronic artwork.
To avoid unnecessary errors you are strongly advised to use the 'spell-check' and 'grammar-check' functions of your word processor.

Article structure

Text

Papers should be organized in the following format: Abstract (which must consist of a

single paragraph only and no sub-headings), Introduction, Materials (or Patients) and Methods, Results and Discussion. Other descriptive headings and sub-headings may be used if appropriate. Every effort should be made to avoid jargon and non-standard abbreviations. Contents of the study should be presented as clearly and as concisely as possible.

Subdivision - numbered sections

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

Introduction

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

Material and methods

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

Results

Results should be clear and concise.

Discussion

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

Conclusions

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

Appendices

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

Essential title page information

• ***Title.*** Concise and informative. Titles are often used in information-retrieval systems. Avoid abbreviations and formulae where possible.
• ***Author names and affiliations.*** Please clearly indicate the given name(s) and family name(s) of each author and check that all names are accurately spelled. You can add your name between parentheses in your own script behind the English transliteration. Present the authors' affiliation addresses (where the actual work was done) below the names. Indicate all affiliations with a lower-case superscript letter immediately after the author's name and in front of the appropriate address. Provide the full postal address of each affiliation, including the country name and, if available, the e-mail address of each author.
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Note shortened form for last page number. e.g., 51–9, and that for more than 6 authors the first 6 should be listed followed by 'et al.' For further details you are referred to 'Uniform Requirements for Manuscripts submitted to Biomedical Journals' (J Am Med Assoc 1997;277:927–34) (see also Samples of Formatted References).

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Appendix H

Main Research Project Interview Topic Guide

Introduction

Points to include:

• Check participants understanding of the purpose of the study, and find out if they have any questions

• Emphasize confidentiality

• Explain that there are no right or wrong answers, we would like to learn from participants' experiences and views

The interviews are semi-structured and iterative, so the content may vary. Questions can be rephrased into a language appropriate for each participant. Questions will include the following:

• Warm up questions, what do you enjoy? What's fun for you?

• Can you tell me about your neuromuscular condition?

Fatigue

Some research suggests that fatigue is a problem for people who have a neuromuscular condition -

• Check understanding of fatigue – how would you describe fatigue?

• Can you tell me about your experiences of fatigue? What does fatigue feel like for you?

• How does it affect you?

• What impact does it have on daily life? E.g., at school, at home, with friends, hobbies

• How does it make you feel? How does it affect your mood?

• How well do other people understand your experiences of fatigue? E.g., parents,

school/college/work, friends.

• What have you found to be the most helpful ways of managing your fatigue?

• Can you tell me about any resources or support that you have found beneficial or would have liked to have been offered?

• Anything else you would like to talk about?

Appendix I

Main Research Project Ethical Approval

From: psychology-ethics <psychology-ethics@bath.ac.uk>
Sent: 03 February 2021 09:17
To: Kiesha Williams <kw733@bath.ac.uk>
Cc: Maria Loades <mel26@bath.ac.uk>
Subject: 20-240

Dear Kiesha

Full title of study: Is fatigue a concern for adolescents with neuromuscular conditions?
PREC reference number: 20-240

On behalf of the Committee, I am pleased to confirm that you have received a favourable ethical opinion for the above proposal from the Psychology Research Ethics Committee.

However please be aware that a researcher (or supervisor in the case of UG or Masters students) is responsible for ensuring full GDPR compliance. Please seek further advice from dataprotection-queries@lists.bath.ac.uk if you have any concerns.
Under current Covid restrictions, if you are proposing lab based or field research involving in-person testing you will also need to get approval from the Psychology Research Restart Group (PRRG) before you can start to gather data. More information can be found here:

https://wiki.bath.ac.uk/display/PC/Psychology+COVID-19+Home

If you intend to display recruitment posters/materials, please ensure you obtain the appropriate permission to do so from those who manage the location(s) you choose.
Please inform PREC about any substantial amendments made to the study if they have ethical implications.

Please make sure you quote your unique PREC code, 20-240, in any future correspondence.

Rebecca Wise
On behalf of Psychology Research Ethics Committee

Appendix J

Main Research Project Participant Quotes

Theme/Subtheme	ID	Quote
1. A constant uncertainty		
- *How fatigue was experienced*	P3	"Fatigue is having literally no energy doing something, being exhausted."
	P4	"I would also describe it as sort of deflating and personally, for me, it feels like I wouldn't even want to reach and pick up the TV remote."
	P6	"It's not very predictable … How do you set up a life which facilitates a level of fatigue which you don't know will or won't be there?"
	P11	"[Fatigue is] very big exhaustion, tiredness."
- *Degree of fatigue*	P3	"The degree of fatigues differ … It can be more, can be less, but … we can't run away from it."
- *Mind and body fatigue*	P4	"It can definitely impact in my uni work because I'll spend obviously my time at work and my brains tired but also if my body is tired, there's no way I'm going to get anything done that evening."
	P5	"Sometimes [I] even find it difficult resting. Since … you've already put in your mind that you have something to do."
	P12	"I just haven't got the mental strength to do it [studying]."
2. Fatigue is multifactorial and can be a 'bit of a cycle'		
- *Interconnected triggers for fatigue*	P1	"Sometimes when I sit for long, I just feel like I'm so so so so tired."
	P2	"When I haven't done anything for a while, I will start to get tired."
	P2	"I have muscle pain which … leads to tired because I couldn't sleep."
	P8	"If I'm very busy at a weekend, so [a sports] tournament … I would feel a lot more fatigued."
	P9	"I won't want to go training because I'll be so tired and I'm sort of there like "Well, why should I go training if it's going to tire myself out even more?"
- *Cycles between fatigue and the neuromuscular condition*	P1	"When I'm stressed with my illness, I just get fatigued."
	P3	"You feel like your body is not normal."
	P4	"In the winter months my muscles feel a lot weaker and so tasks take more energy … that would definitely lead to me feeling more fatigued at the end of the day."

3. Fatigue interferes with different areas of life		
- *Day-to-day tasks*	P4	“It could affect my work.”
	P5	“I can't work because I'm feeling tired.”
- *Thinking abilities*	P8	“The best way to put it is when I'm struggling to find the right words.”
	P12	“When I’m tired … my speech messes up … sometimes I can’t tell I’m doing it, but I slur a lot without realising.”
- *Personal and social impact*	P5	“Maybe you might want to maybe catch up with your friends. You can't do it. You feel like you’re irritating them.”
	P8	“If I'm struggling to get words out, it's kind of embarrassing.” “It just kind of felt like weekdays were sort of off limits, you know you can't do anything with your mates in the week”
- *Mood*	P5	“It makes me feel so bad because you miss out on a lot … It really affects my mood because sometimes I feel as if I’m not normal … I feel different since I’m not able to … do my tasks in the right way, I’m not able to do a lot of things.”
	P11	“I can get frustrated at myself, I think because I'm not doing so much … because I’ve got low energy, my mood can sometimes be quite low.”
	P6	“It limits how much I can do. It limits the quality of how much I can do and it's a reminder of your disability as well, which mental health wise isn’t wonderful, like when you’re feeling rubbish and your body won’t just do normal stuff, that sucks.”
4. It’s tricky to manage fatigue		
- *Different things work at different times for different people*	P2	“My mind wasn’t really on anything else, just wanting to rest, and the problem is when you wake up from naps and they make you even more tired”
	P3	“If you’re feeling exhausted you can … make it a little easier by doing a very light walk.”
	P4	“I guess it depends how tired I am. Sometimes … I just need to go to bed and start fresh in the morning, reset that battery back to 100%.”
	P6	“I still sometimes get it wrong, like I'm very good at reading my body, not always good at listening to it but I kind of usually know when sometimes an hour [of rest] doesn’t help, and you’re like “oh well, that’s an hour of my life wasted” as my body still feels tired.”
	P11	“I think if you keep yourself active, you have a bit more energy to begin with.”

- *Changing plans*	P3	"You are exhausted so at times it affects attending activities."
	P4	"I need to … stick to a schedule with my routines so that I know where I'm going to need the most energy in the day, I know where I'm going to need to be the most present."
	P5	"Maybe I may have an appointment with my friends. Maybe we planned on doing something, but now when that time comes, I'm unable to do anything. I'm unable to. I just feel like staying in bed."
	P6	"[I] constantly have like a mental calculation in my head of how much I can push, how much can I do that? I plan everything pretty much … I just factor in rest breaks."
- *Push through*	P6	"I usually push through, but then that makes it so so much worse."
	P10	"When I do feel fatigue, I just push myself a lot."
	P12	"When the fatigue hits me, I still try and push through it … it takes longer to recover."

5. Mixed responses to fatigue (internal and external)

- *Normalising*	P1	"When I'm fatigued, I just tend to feel like it's just a normal day for me."
	P3	"It's like I'm used to it."
	P10	"My normal is always being tired."
- *Minimising*	P6	"I say I get tired, but I don't like using the word fatigue because I know that's a separate medical thing, but also I do get tired."
- *Mixed feelings about disclosing fatigue and being treated differently*	P1	"When around people you just tend to be strong and you just pretend that you're not fatigued, but in real sense, you [are] so so, so much fatigued."
	P2	"I don't tell them about it because I don't want to be treated any differently."
	P9	"My form tutor … she does try and help me quite a lot …with tiredness … they don't really pick on me during the last lessons because they know that I'm pretty tired."
- *People try to be supportive but it's hard to understand*	P4	"They can say they understand it, but until you're actually living someone's experience, I don't think you'll ever truly, truly understand it … obviously they do try because … they want to show some empathy."

	P5	“They don't know if it [fatigue] exists or not…. they're like ‘what's that, is it even real?”
	P6	[People ask] “Is there anything we can do to stop you having bad days?” … the idea that if I did rest a set amount or treated myself like an able body then I’d wake up the next day feeling completely fine, and … that’s not how it works. The amount of reward you can get is minimal.”
	P7	“They assume that everyone is the same… they kind of wrap me up in cotton wool sometimes and … I'm absolutely fine.”
	P12	“Trying to explain to other people when I don’t even understand it … it's hard.”
- *Fatigue is rarely discussed in healthcare*	P4	“I don't think professionals focus on it [fatigue] as much … Every time I've said that I’m tired, they're like, ‘oh, it must be your lungs. It must be this.’ They don't actually think about the impact … and so I do feel like it has been ignored in the past.”
	P5	“She just advised me to take lots of rest. I don't think she understands what fatigue is.”
	P6	“It would be quite nice to have it recognised as a symptom … That would make the mental strain a bit easier, if like it was more accepted.”
	P11	“I don't think they have really talked about fatigue … they've just talked about managing pain.”

Appendix K

Main Research Project Participant Information Forms

Parent Information Sheet

Your child is being invited to take part in a research study. Before deciding with them whether they would like to take part, both you and your child should understand what the research is about and what they will be asked to do. Please read this information sheet and let the research team know if you have any questions.

Please click this link if you would like to download and read the information in a PDF:
Downloadable Parent Information Sheet

Why are we doing this research?
Research has shown that young people who experience physical health conditions, such as cancer and physical disabilities, can experience 'fatigue' which has been described as feelings of 'exhaustion' and a 'lack of energy'. Research has also looked at this in adults who have specific neuromuscular conditions. We don't know if this also affects young people across different neuromuscular conditions. We would like to find this out and look at whether it impacts on other areas of life, such as sleep, school and social activities. In the future, this will hopefully help us to know more about how we can support children and young people with neuromuscular conditions.

To do this research, we are using 7 questionnaires. These measure fatigue, sleep quality, school/social functioning, mood, physical health, quality of life, and beliefs and behaviours. We would like young people to fill these in.

As part of the study, we will also contact participants to invite them to take part in a follow-up interview, looking at the experiences of fatigue.

Why has my child been chosen? Your child has been chosen to take part in this study because they are aged between 10 and 24 years and have a neuromuscular condition.

Does my child have to take part? No, taking part in this study is voluntary. If your child does decide to take part, they can stop completing the questionnaires at any point once they have started. The questionnaires will be online and if they choose to close the web browser all their information will be deleted from the study. They will not have to give any reason for not taking part or for deciding to exit the survey.

Once they have finished the questionnaires, they will be asked if they are happy to submit their answers:
• If they tick the "no" box, their answers will not be included in the study and their data will be withdrawn and deleted.
• If they tick the "yes" box, their answers will be used in the study and they will no longer be able to withdraw their data from the study.

What will my child be asked to do if we decide to take part?
• If you and your child would like to find out more about the research, you can contact the researcher on: kw733@bath.ac.uk

• If they would like to take part, you and your child can click next at the end of this page – you will then both be invited to complete consent forms and your child will be able to continue to complete the questionnaires.
• If your child is under 16, you will be asked to fill in a form to say that you agree for them to take part. If they are aged 16 or over, they can fill this form in themselves. The consent forms will ask for an email address - this is an invitation to be entered into a prize draw for the chance to win one of three £20 gift vouchers. The email address will be stored securely and separate from the questionnaire responses.
• Your child will then be asked to fill in their demographics and then seven questionnaires. We expect this to take between 20 - 30 minutes.
• When they have finished the questionnaires, they will be shown some more information about the research. It would be helpful for you to read this information through with your child.
• It is important that your child feels comfortable reading and understanding English so that they can answer questions independently. You can help your child to read any questions they do not understand but you cannot help them answer.
• After completing the questionnaires, the researcher will contact you to arrange to meet with the young person using Microsoft Teams for a follow up interview. This will involve the researcher asking the young person some questions about their experiences of fatigue. Interviews are expected to last around 20-30 minutes. The interviews will be recorded using Microsoft Teams - if your child does not wish to be video-recorded then they can turn their camera off and the interview will be audio recorded only.

Do I need to worry about my child taking part? The study will ask your child to answer questions about their thoughts, feelings, and experiences. Answering questions on these things could be upsetting. If your child feels upset when taking part, they can stop at any time. There are no right or wrong answers and the responses they give will not affect their current care or treatment. If you notice that your child does feel sad or worried during or after taking part, it is important to talk to them about this. It may also be helpful to ask if they would like to speak to their GP or school nurse/counsellor about how they are feeling. Researchers will not inform your child's GP if they take part in this study and so it may be helpful to let your child's GP know about the research if they do experience any distress following participation.

Are there any benefits to taking part? If your child does decide to take part in this study, their responses will hopefully help us to understand whether fatigue is a problem for those living with a neuromuscular condition, and the impact of this on areas of their life. We hope that this will lead to better support for young people in the future. Taking part also means your child will be entered into an optional prize draw for a chance to win a £20 gift voucher.

Will my child's answers be confidential? The information your child provides will be confidential and anonymous. Everyone who takes part will be assigned a unique number, and the answers they provide will be stored with that number, rather than their name. This means that only the research team will be able to link their answers to them.

As the study is anonymous, the researchers will also not know the names of who has taken part. This means that once your child has submitted their questionnaires, they will not be able to withdraw their information. At the end of the questionnaires, they will be

asked if they are happy to submit their answers. If they answer ‘no’, all of their information will be withdrawn and deleted.

After the interviews, the audio/video recordings will be transcribed by the researcher. The transcriptions will remain anonymous and will be stored under the same unique number as the survey. Names will not be stored with the data. What will happen to my child’s answers after they take part? Your child’s answers to the questionnaires will be kept on a secure computer file and kept anonymously (with a number, not their name). They will be stored at the University of Bath for 10 years. After this, their anonymised answers will be securely stored at the University of Bath Archives. This is where previously collected information is stored so that it can be looked at by researchers if needed. The findings will be used to help us to understand the experiences of fatigue for young people with neuromuscular conditions. We plan to publish the findings of this study in professional journals and to share the findings at conferences and through professional networks. If you would like a summary of the study results after it has finished, you can ask the researcher for this after your child has taken part. Any results from the study that are published will not include any identifiable information. In the longer term, we hope this information will be used to support young people with neuromuscular conditions. We will also consider making the data available for other researchers to access via the Open Science framework but will ensure that your child's anonymity is not compromised in doing so.

What if I have any questions or if I have a problem? If you have any questions about this study, please contact the research team. Firstly, you should contact the main researcher (Kiesha Williams). If needed, you can also contact the academic or field supervisors. They will aim to get back to you within two weeks.
Main Researcher – Kiesha Williams Email: kw733@bath.ac.uk
Academic Supervisor – Dr Maria Loades Email: m.e.loades@bath.ac.uk
Field Supervisor – Dr Sadie Thomas-Unsworth Email: sadie.thomas-unsworth@uhbw.nhs.uk

Who is organising this study? This study is being completed by a doctoral student at the University of Bath. It is closely supervised by members of staff in the Department of Psychology at the University of Bath. Who has approved this study? This study has been given ethical approval by the Department of Psychology Research Ethics Committee at the University of Bath (Ref: 20-240) If you are unhappy with the way the research is carried out and would like to complain, you can do this by contacting the Department of Psychology Research Ethics Committee at the University of Bath - email: psychology-ethics@bath.ac.uk Thank you for taking the time to read this information.

Information Sheet – Age 10 – 15

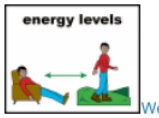
UNIVERSITY OF BATH

Fatigue in Neuromuscular Conditions
Information for young people aged 10 - 15 years

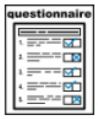
We would like to invite you to take part in a research study. Before you decide whether you would like to take part, you and your parent or carer should understand what you will be asked to do. Please read this information sheet and let the research team know if you have any questions.

Why are we doing this research?

- We know that young people with some health conditions can experience 'fatigue' - this is a feeling of exhaustion or lack of energy. This has also been investigated in adults who have specific neuromuscular conditions.

- 

We don't know if fatigue also affects young people with different neuromuscular conditions, and we would like to find this out. We also want to understand whether fatigue affects other areas of life, such as school/work, sleep, and friendships. In the future, this will hopefully help us to know more about how we can support others with neuromuscular conditions.

- 

To look at these things, we would like young people to fill in 7 questionnaires and complete a follow-up interview with the researcher. These will ask questions about fatigue, sleep, school, how you feel, physical health, quality of life, and thoughts or behaviours.

Why have I been chosen? 

We would like to work with young people with neuromuscular conditions, who live in the UK, and are aged between 10 and 24 years. To take part you will be asked to fill out online questionnaires and meet with the researcher using Microsoft Teams who will ask you some questions about fatigue.

Do I have to take part?

No, you do not have to take part. ***You can decide if you would like to take part together with your parent/carer.***

If you decide to take part, you can stop doing the questionnaires at any time before you finish. You can close the browser at any time before you finish, and all your information will be deleted. You do not have to tell anyone why you do not want to take part or why you decided not to finish the questionnaires.

If you do finish the questionnaires, at the end of the study you will be asked if you are happy for your answers to be sent to the researchers running the study:

 Ticking the "no" box means you would not like your answers to be included in the study. This means your answers will be deleted.

Ticking the "yes" box means that you would like your answers to be used in the study. Ticking this box means that your answers will not be deleted. You will not be able to delete your answers after this.

If I decide to take part, what will I be asked to do?

1. 

Both you and your parent/carer will read through this **information sheet.** If you decide to take part, you can click next at the end of this online page. If you have any questions you can email the researcher – Kiesha Williams on kw733@bath.ac.uk

2. 

You and your parent/carer will then be asked to fill in a **consent form** so that we know you have agreed to take part in the study.

3. 

After this, you will be asked to fill out some information about yourself (e.g. age and gender), and then the seven **questionnaires**. This should take you around 20 - 30 minutes in total.

4. 

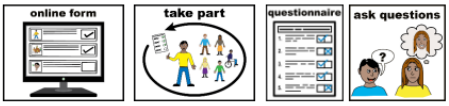
When you have finished the questionnaires, you will be shown some more information about the research and **asked if you are happy for your answers to be used** in the study.

5. 

The researcher will then arrange to meet with you online, using Microsoft Teams. They will ask you some more **questions about fatigue**. You will have a chance to ask any questions before starting. This meeting will last 20-30 minutes.

6. 

If you and your parent/carer have agreed and entered an email address, you will be entered into a **prize draw** after taking part. The prize draw is for the chance to win a £20 gift voucher. Email addresses will be stored securely and separate from your answers.



Do I need to worry about taking part?

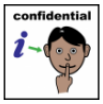
This study will ask you questions about your thoughts, feelings, and experiences. Answering questions on these things could be upsetting. If you feel upset when taking part, you can stop. There are no right or wrong answers and the responses you give will not affect your current care or treatment.

If you feel sad or worried during or after taking part, it is important to talk to someone about this. You can tell your family or speak to your GP, school nurse/counsellor, teacher, or support services. There is some additional information at the end of this page on where to access support.

Are there any benefits to taking part?

If you decide to take part in this study, your responses will hopefully help us to understand the experience of living with a neuromuscular condition. We hope that this will lead to better support for young people in the future.

After taking part in the study you can also choose to be entered into a prize draw for a chance to win one of three £20 gift vouchers.



Will my answers be confidential?

The information you provide will be confidential. This means that it will be kept private. The responses you give will be assigned a unique number – all the information you provide will

be stored under that number, rather than your name. This means that only the research team will be able to link your answers to you.

What will happen to my answers after I take part?

Your answers to the questionnaires will be kept on a secure computer file and stored privately, with a unique number – not with your name. This will be stored at the University of Bath for 10 years. After this, your answers will be securely stored at the University of Bath Archives. This is a place where information that previously been collected is stored so that it can be looked at by researchers if needed.

If you would like a summary of the study results after it has finished, you can ask the researcher for this after taking part. Any results from the study that are published will not include any identifiable information.

What if I have any questions or if I have a problem?

If you have any questions about this study, please contact the research team.

My name is Kiesha Williams, I am the main researcher for this project. You can contact me on: kw733@bath.ac.uk

If needed, you can also contact the academic or field supervisors. They will aim to get back to you within two weeks.

Main Researcher	**Kiesha Williams** kw733@bath.ac.uk
Academic Supervisor	Dr Maria Loades m.e.loades@bath.ac.uk
Field Supervisor	Dr Sadie Thomas-Unsworth sadie.thomas-unsworth@uhbw.nhs.uk

Who is organising this study?

This study is being completed by a doctoral student at the University of Bath. It is closely supervised by members of staff in the Department of Psychology at the University of Bath.

Who has approved this study?

This study has been given ethical approval by the Department of Psychology Research Ethics Committee at the University of Bath (Ref: 20-240)

If you are unhappy with the way the research is carried out and would like to complain, you can do this by contacting the Department of Psychology Research Ethics Committee at the University of Bath - email: psychology-ethics@bath.ac.uk

Feeling down or worried?

If you feel upset or worried after taking part, you may find it helpful to talk to someone. This could be a GP, parent, or school nurse/counsellor/teacher. These people can help you to get support for how you are feeling.

Some other places that offer support and more information about mental health difficulties include:

YoungMinds - https://youngminds.org.uk/

YOUNGMiNDS

Childline - www.childline.org.uk or call for free on 0800 1111

ChildLine 0800 1111

Thank you for taking the time to read this information.

If you have any questions, or would prefer to fill in paper copies of the questionnaires, please contact Kiesha Williams kw733@bath.ac.uk

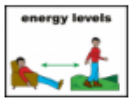
Age 16 – 24 Information Sheet

Fatigue in Neuromuscular Conditions
Information for young people aged 16 - 24 years

We would like to invite you to take part in a research study. Before you decide whether you would like to take part, you should understand what the research is about and what you will be asked to do. Please read this information sheet and let the research team know if you have any questions.

Why are we doing this research?

- **Research has shown that young people with some long-term health conditions can experience 'fatigue' which has been described as feelings of 'exhaustion' and a 'lack of energy'. Research has also looked at this in adults who have specific neuromuscular conditions.**
- **We don't know if this is also something that affects young people across different neuromuscular conditions.**
- **We would like to find this out and look at whether it impacts on other areas of life, such as sleep and social activities.**
- **In the future, this will hopefully help us to know more about how we can support children and young people with neuromuscular conditions.**



To do this research, we are using 7 questionnaires. These ask questions about fatigue, sleep quality, school/social functioning, mood, physical health, quality of life, and beliefs and behaviours. We would like young people to fill these in.

The researcher will also meet with you for a follow-up interview using Microsoft Teams. The researcher will ask you some more questions about fatigue. This meeting will take 20-30 minutes.

Why have I been chosen?

We would like to work with young people with neuromuscular conditions in the UK, aged between 10 and 24 years, who are able to fill out online questionnaires and answer questions in English.

Do I have to take part?

No, taking part in this study is voluntary – this means that you do not have to take part.

If you decide to take part, you can stop doing the questionnaires or interview at any time before you finish. The questionnaires will be online so if you choose to stop taking part before you finish, you can close the browser and all your information will be deleted. You do not have to tell anyone why you do not want to take part or why you decided not to finish the questionnaires.

If you do finish the questionnaires, at the end of the study you will be asked if you are happy for your answers to be sent to the researchers running the study:

-  Ticking the "no" box means you would not like your answers to be included in the study. This means your answers will be deleted.

-  Ticking the "yes" box means that you would like your answers to be used in the study. Ticking this box means that your answers will not be deleted. You will not be able to delete your answers after this.

If I decide to take part, what will I be asked to do?

1. If you decide to take part, you can **click next** at the end of this online page. If you have any questions, you can email the researcher, Kiesha Williams - kw733@bath.ac.uk

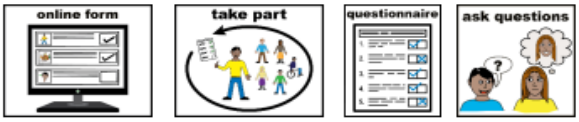
2. After reading this sheet, you will be asked to fill in a **consent form** so that we know you have agreed to take part in the study.

3. You will then be asked to fill in some information about yourself (e.g. age and gender), and then the seven **questionnaires**. This should take you around 20 - 30 minutes in total.

4. When you have finished the questionnaires, you will be shown some more information about the research and **asked if you are happy for your answers to be used in the study**.

5. The researcher will contact you to arrange a time to **meet online, using Microsoft Teams** and ask you some more questions about fatigue. This will take 20-30 minutes. In the meeting you will have the chance to ask questions before starting.

6. If you have agreed and entered your email address at the beginning of the study, you will be entered into a prize draw after taking part. The **prize draw** is for the chance to win a £20 gift voucher. Email addresses will be stored securely and separate from your questionnaire responses.



Do I need to worry about taking part?

This study will ask you questions about your thoughts, feelings, and experiences. Answering questions on these things could be upsetting. If you feel upset when taking part, you can stop. There are no right or wrong answers and the responses you give will not affect your current care or treatment.

If you feel sad or worried during or after taking part, it is important to talk to someone about this. You can tell your family or speak to your GP, school nurse/counsellor, teacher, or support services. There is some additional information at the end of this sheet on where to access support.

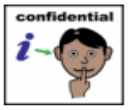
Are there any benefits to taking part? 

If you decide to take part in this study, your responses will hopefully help us to understand the experience of living with a neuromuscular condition. We hope that this will lead to better support for young people in the future.

After taking part in the study you can also choose to be entered into a prize draw for a chance to win one of three £20 gift vouchers.

Will my answers be confidential?

The information you provide will be confidential. This means that it will be kept private. The responses you give will be assigned a unique number – all the information you provide will be stored under that number, rather than your name. This means that only the research team will be able to link your answers to you.



What will happen to my answers after I take part?

After the interview, the researcher will write out your responses. Once written down, the recording will be deleted and only the written responses and questionnaire answers will be stored on a secure computer file and stored privately, with a number – not with your name. This will be stored at the University of Bath for 10 years. After this, your answers will be

securely stored at the University of Bath Archives. This is a place where information that previously been collected is stored so that it can be looked at by researchers if needed.

If you would like a summary of the study results after it has finished, you can ask the researcher for this after taking part. Any results from the study that are published will not include any identifiable information.

What if I have any questions or if I have a problem?

If you have any questions about this study, please contact the research team.

My name is Kiesha Williams, I am the main researcher for this project. You can contact me on: kw733@bath.ac.uk

If needed, you can also contact the academic or field supervisors. They will aim to get back to you within two weeks.

Main Researcher	**Kiesha Williams** **kw733@bath.ac.uk**
Academic Supervisor	Dr Maria Loades m.e.loades@bath.ac.uk
Field Supervisor	Dr Sadie Thomas-Unsworth sadie.thomas-unsworth@uhbw.nhs.uk

Who is organising this study?

This study is being completed by a doctoral student at the University of Bath. It is closely supervised by members of staff in the Department of Psychology at the University of Bath.

Who has approved this study?

This study has been given ethical approval by the Department of Psychology Research Ethics Committee at the University of Bath (Ref: 20-240).

If you are unhappy with the way the research is carried out and would like to complain, you can do this by contacting the Department of Psychology Research Ethics Committee at the University of Bath - email: psychology-ethics@bath.ac.uk

Feeling down or worried?

If you feel upset or worried after taking part in this project, you may find it helpful to talk to someone. This could be a GP, parent, or school nurse/counsellor/teacher. These people can help you to get support for how you are feeling.

Some other places that offer support and more information about mental health difficulties include:

YoungMinds - https://youngminds.org.uk

YOUNGMiNDS

Childline - www.childline.org.uk or call free on 0800 1111

ChildLine 0800 1111

Thank you for taking the time to read this information.

If you have any questions, or would prefer paper copies of the questionnaires, please contact Kiesha Williams kw733@bath.ac.uk

Appendix L

Main Research Project Consent Forms

Consent Form for Parents and Carers
You should have read the Parent Information Sheet which will have explained the research to you and your child.

Please read through the following statements and select **yes** to indicate that you understand and agree with the statements and are happy for your child to take part in this study. If you have any questions whilst completing this form please contact the main researcher by emailing kw733@bath.ac.uk

1. I confirm that I have read and understand the information sheet for the above project

2. I have had the time and opportunity to consider the information.

3. I have had the opportunity to ask questions via email and if I have asked questions I am happy with the answers

4. I have been given enough information about the project to make an informed decision about whether my child would like to be involved in the project

5. I understand that my child's participation is voluntary, and they are free to withdraw at any time whilst filling in questionnaires or during the interview, without giving a reason, and their data will be withdrawn

6. I understand that because the information my child provides within the study is anonymous, at the end of the study my child will be asked if they are happy to submit their data, following this they will no longer be able to withdraw from the study. If they say they are not happy to submit their data, it will be withdrawn from the study

7. I agree to the researcher using information and quotations provided by my child within the report of the study and I understand that all data and quotations will be made anonymous

8. I understand that the interview with my child will take place via Microsoft Teams and will be video and audio recorded, and I agree to this. *Note - if you do not agree to a video recording, the interviewer will ask your child to turn their camera off during the interview so that only audio is recorded.*

9. I understand that researchers from the University of Bath may need to look at the anonymous data to make sure that the study is being completed properly. I give permission for my child's anonymous data to be looked at if needed

10. I give permission for the information my child provides to be stored anonymously and securely at the University of Bath for the duration of 10 years after the study is completed and then archived in the University of Bath data archive with no identifiable data

11. I understand that researchers from outside the University of Bath may request to have access to the anonymised research data once it has been archived. I

understand that the data that they have access to will not include any information that will allow my child to be identified

12. I understand that the information my child provides will be anonymous and confidential. No link will be made between their name or other identifying information and the information my child provides.

13. I hereby fully and freely consent to my child's involvement in this study
I understand the nature and purpose of their involvement in this study. These have been communicated to me on the information sheet accompanying this form or via email with the researcher.

I understand that the information my child provides will be **anonymous and confidential**. No link will be made between their name or other identifying information and the information my child provides.

I have read and understood the above statements and agree for my child to take part in the above named study.

- ○ Yes
- ○ No

If yes, please enter your email address below.

This email address will be used to contact you to arrange a time to complete the follow up interview with the young person. We will also use this email address to send out the gift voucher if you agree for your child to be entered into the prize draw and they win.

I agree for my child to be entered into a prize draw for the chance to win one of three £20 love to shop gift vouchers.

Please tick this box for your child to be entered into the prize draw. If won, the gift voucher will be sent to the above email address.

- ○ Yes
- ○ No

Name (please print):

Signature

SIGN HERE

clear
Date:

Thank you for agreeing for your child to take part in the study. The next page will ask your child whether they agree to the study. They will then be asked some questions about their background and medical condition - you may want to help your child to complete these questions.

If you have any concerns related to your child's participation in this study please direct them to the Department of Psychology Research Ethics Committee email: psychology-ethics@bath.ac.uk (Ref: 20-240)

Young Person, aged 10 - 15, Assent Form

Please select **yes** if you agree with the following questions and **no** is you do not agree.

Have you read information with your parent or carer that explains the project to you?

- Yes
- No

Do you understand what this project is about?

- Yes
- No

Have you asked all the questions you would like to ask? Please email the researcher or speak to your parent/carer if you do have any questions.

- Yes
- No

If you have asked questions, have you had your questions answered in a way that you understand?

- Yes
- No

Do you understand that the research involves answering online questionnaires and meeting with the researcher online where they will ask you some follow up questions?

- Yes
- No

Do you understand that you can stop filling out the questionnaires at any time before you have submitted your answers and that closing the webpage will mean your data is deleted?

- Yes
- No

Do you understand that because the answers you give will be anonymous, once you have submitted the answers you will no longer be able to withdraw your data?

- Yes
- No

Are you happy to take part?

- Yes
- No

Are you and your parent or carer happy for you to be entered into a prize draw for the chance to win one of three £20 love to shop gift vouchers? If you win, this voucher will be sent to the email address provided by your parent or carer.

- Yes
- No

Thank you for your help!

If you have any concerns related to your involvement in this study please direct them to the Department of Psychology Research Ethics Committee email: psychology-ethics@bath.ac.uk (Ref: 20-240)

Consent Form for Young People, aged 16 - 24

You should had read the Information Sheet for young people aged 16-24 which will have explained the research to you. Please read through the following statements and select **yes** to indicate that you understand and agree with the statements. If you have any questions whilst completing this form, please contact the main researcher by emailing kw733@bath.ac.uk.

1. I confirm that I have read and understand the information sheet for the above project

2. I have had the time and opportunity to consider the information

3. I have had the opportunity to ask questions via email and, if I have asked questions, I am happy with the answers

4. I have been given enough information about the project to make an informed decision about whether I would like to take part

5. I understand that participation is voluntary and that I am free to withdraw at any time whilst filling in online questionnaires or during the interview. I do not have to give a reason for this and my data will be withdrawn

6. I understand that because the information I provide is anonymous, at the end of the study I will be asked if I am happy to submit my data. Following this, I will no longer be able to withdraw from the study. If I click that I am not happy to submit data, the responses will be withdrawn from the study.

7. I agree to the researcher using information and quotations provided by me within the report of this study. I understand that all data and quotations will be made anonymous.

8. I understand that the interview will take place via Microsoft Teams and will be video and audio recorded, and I agree to this. Note - if you do not agree to a video recording, the interviewer will ask you to turn off your camera during the interview so that only audio is recorded.

9. I understand that researchers from the University of Bath may need to look at the anonymous data to make sure that the study is being completed properly. I give

permission for my anonymous data to be looked at, if needed.

10. I give permission for the information I provide to be stored anonymously and securely at the University of Bath for the duration of 10 years after the study is completed and then archived in the University of Bath data archive with no identifiable data.

11. I understand that researchers from outside the University of Bath may request to have access to the anonymised research data once it has been archived. I understand that the data that they have access to will not include any information that will allow me to be identified.

12. I understand that the information I provide will be anonymous and confidential. No link will be made between my name or other identifying information and the information I provide.

13. I hereby fully and freely consent to my involvement in this study

I understand why I am participating in this study and have read the information sheet with this form.

I understand that the information I provide will be **anonymous and confidential.** No link will be made between my name or other identifying information and the information I provide.

I have read and understood the above statements and agree to take part in the above named study.

- ○ Yes
- ○ No

If yes, please enter your email address below.

This email address will be used to contact you to arrange a time to complete the follow up interview. We will also use this email address to send out the gift voucher if you agree to be entered into the prize draw and win.

I agree to be entered in a prize draw for the chance to win one of three £20 love to shop gift vouchers.

Note - if you win, the voucher will be sent to the email address provided above.

- ○ Yes

- No

Name (please print):

Signature

SIGN HERE

clear

Date:

If you have any concerns related to your involvement in this study please direct them to the Department of Psychology Research Ethics Committee email: psychology-ethics@bath.ac.uk

Appendix M

Main Research Project Debrief Form

Debrief Form
Fatigue and Neuromuscular Conditions

The aim of this study is to understand whether fatigue is something that young people with neuromuscular conditions struggle with, and if so, how common it is.

You have been asked some questions about fatigue, sleep, mood, health and quality of life, social activities, and thoughts and behaviours. Research with young people with long term health conditions shows us that fatigue can be a problem and may impact on these other areas of life - but we don't know if this is also true for those with neuromuscular conditions. We predict that fatigue will be common and that it will have the biggest impact on mood and quality of life.

Why is this important?
We want to understand this as fatigue may be under-recognised by clinicians and services. Recent research has also shown that those with a specific type of neuromuscular disorder can experience high levels of fatigue. We wanted to investigate whether this is similar or different for those with other neuromuscular conditions.

What next?
The researcher (Kiesha Williams, kw733@bath.ac.uk) will contact you on the email address you provided at the beginning of the study to arrange a follow-up interview using Microsoft Teams.

Are you happy for your answers to be sent to the researchers running the study

☒ No - this means you do not want your answers to be in the study. Your answers will be deleted.

☑ Yes - this means you would like your answers to be used in the study. Ticking this means your answers will be sent to the researcher and you won't be able to delete your answers anymore.

Thank you for participating in our study.

If you have found this research upsetting or distressing in any way, it is important to talk to someone about this. Below is a list of sources that can provide support:

- **Childline** - www.childline.org.uk or call for free on 0800 1111
- **YoungMinds** - https://youngminds.org.uk/
- **Your GP**
- **Your school nurse or counsellor**

If you would like to receive a summary of the study's main findings please enter your email address below:

If you have any questions about the project, please contact Kiesha Williams - kw733@bath.ac.uk

Appendix N

Main Research Project Online Questionnaires

Inclusion criteria

Thank you for agreeing to take part in this study. Over the next pages you will be shown questions about yourself, please read each question carefully.

Are you aged 10 - 24 years old?

Yes

No

Do you have a neuromuscular condition?

Yes

No

Do you live in the UK?

Yes

No

Are you able to complete questionnaires and a follow up interview online, in English?

Yes

No

Demographics

The next questions will ask for some more information about you. If you are unsure, please ask your parent/carer to help you.

What is your age (in years)?

What is your gender?
Male
Female
Transgender
Prefer not to say

What is your ethnic group? Choose one option that best describes your ethnic group or background

White:
English / Welsh / Scottish / Northern Irish / British
Irish
Gypsy or Irish Traveller
Any other White background, please describe

Asian / Asian British:
Indian
Pakistani
Bangladeshi
Chinese
Any other Asian background, please describe

Black / African / Caribbean / Black British:

African
Caribbean
Any other Black / African / Caribbean background, please describe

Mixed / Multiple ethnic groups:

White and Black Caribbean
White and Black African
White and Asian
Any other Mixed/ Multiple ethnic background, please describe

Other ethnic group:

Arab
Any other ethnic group, please describe

What neuromuscular condition do you have?

Duchenne muscular dystrophy
Becker muscular dystrophy
Bethlem myopathy
Congenital muscular dystrophy
Congenital myotonic dystrophy

Emery-Dreifuss muscular dystrophy
Facioscapulohumeral muscular dystrophy
Limb-girdle muscular dystrophy
Myotonic dystrophy
Myofibrillar myopathy
Spinal muscular atrophy
Congenital myopathy
Charcot-Marie-Tooth disease
Congenital myasthenic syndromes
Other, please give details:

Do you have any other medical conditions or diagnoses?
No
Yes, please give brief details

Are you currently taking any medication?
No
Yes, please give brief details

Are you currently a wheelchair user?
Yes
No

If yes, what percentage of time did you use a wheelchair to get round this week?

0-25%
25-50%
50-75%
75-100%

Where did you hear about this study?

UK Myotonic Dystrophy Registry
UK FSHD Registry
Children's Hospice
Social Media
Action Duchenne
Muscular Dystrophy UK
Other, please give brief details

The following questionnaires were included in the online survey:

- Chalder Fatigue Questionnaire
- Revised Children's Anxiety and Depression Scale-25
- Short-Form 36 Physical Functioning Scale
- Cognitive Behavioural Responses Questionnaire – Short Version
- The School and Social Adjustment Scale
- Kidscreen-27
- Adolescent Sleep-Wake Scale-Short Version

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