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What explains high life satisfaction in men living with Duchenne Muscular Dystrophy? A preliminary study to inform psychological intervention.

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Running title: Life satisfaction in men with DMD

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Abstract

Introduction: There are increasing numbers of men with DMD (mDMD). For those struggling to live with the condition, psychological interventions may be helpful. However, it is unclear how these should be tailored for mDMD. To inform intervention, this study assessed whether 2 well-validated psychological models (Leventhal Self-Regulatory Model; Psychological Flexibility Model) could explain variation in life satisfaction (LS).

Methods: Sixteen mDMD, aged 18-43, completed an online survey comprised of questionnaire measures of LS, mood, and both psychological models: Illness Perceptions (Leventhal Self-Regulatory Model); engagement in meaningful activity, and acceptance/awareness of difficult thoughts and feelings (Psychological Flexibility Model). A median split enabled comparison of high and low LS groups.

Results: Those with higher LS were characterized by the ability to undertake personally meaningful activity in acceptance of difficult thoughts and feelings.

Conclusion: Results supported the Psychological Flexibility Model. However, methodological limitations mean that these findings should be considered preliminary.

duchenne muscular dystrophy; psychology; Acceptance and Commitment Therapy; psychological flexibility; clinical psychology; mood

Improved symptomatic treatments have resulted in increased life expectancy in men with DMD (mDMD); nowadays 60% will live into their 20s and beyond (1). Thus, there is a growing population of mDMD whose needs must be met by health services (2). Studies of well-being [measuring quality of life or life satisfaction (LS)] in mDMD demonstrate that, although physical aspects are reduced, psychosocial aspects remain relatively intact (3, 4). However, there are some exceptions (5), and reflecting this, best practice guidelines recommend that multi-disciplinary care teams should be equipped to manage problematic mood (depression/anxiety etc.) and well-being (6). An improved understanding of the psychological processes affecting LS in mDMD can inform such interventions (7). A small number of qualitative studies have touched on psychological processes. These suggest that mDMD are often frustrated by barriers to adult relationships and other personally meaningful activity, such as vocational pursuits and leisure activity (3, 8). This implies that those engaged in personally meaningful activity may experience better LS; this has yet to be examined in detail, and other explanations are also viable. However, to date, there has been no direct study of the influence of psychological processes on outcomes in this population.

This study investigated the explanatory value of 2 well-researched candidate psychological models for explaining LS in mDMD. First, the Psychological Flexibility Model, which posits that LS is best explained by one's ability to enact personally meaningful activity, such as socializing and working, in acceptance of the difficult thoughts and emotions which naturally arise as one lives with illness (9). Second, the Leventhal Self-Regulatory Model, which theorizes that one's illness perceptions (beliefs about how long a condition will last, its consequences, whether it can be cured) influence the methods used to cope with illness and that this explains variations in LS (10, 11). Based on our work with adult-onset muscle disorders (7, 12) and the

aforementioned qualitative research (3, 8), we hypothesized that the Psychological Flexibility Model would better explain variation in the LS of mDMD.

Methods

Detailed description of the methods and measures can be found in (12) . Data were captured using an online survey hosted by Muscular Dystrophy UK, which was open to adults with all forms of muscular dystrophy. The data presented here are from a previously unanalyzed subsample of mDMD participants. Participants applied inclusion and exclusion criteria for themselves. Inclusion criteria included a diagnosis of muscular dystrophy of duration greater than 6 months, and aged between 18 and 75 years. Exclusion criteria included cognitive impairment and participation in treatment studies. Subjects then completed a consent form and verification questions. Validated questionnaire measures then captured: LS (Satisfaction With Life Scale; SWLS) (13); anxiety (Generalized Anxiety Disorder 7- Item Scale; GAD-7)(14); and depression (Patient health Questionnaire; PHQ-9)(15). Illness perceptions were recorded using the 8 single-item domains of the Brief Illness Perception Questionnaire (B-IPQ)(16). Here, higher scores indicate a stronger belief that DMD has severe consequences (consequences), has many symptoms (identity), is chronic (timeline acute/chronic), can be controlled by behavior (personal control) or treatment (treatment control), is understandable (illness coherence), causes concern (concern), or causes distress (emotional representation). Facets of psychological flexibility were measured using 3 questionnaires; 1) the Engaged Living Scale (ELS)(17) captured valued-living, the extent of engagement in personally meaningful activity; 2) the Acceptance and Action Questionnaire (AAQ)(18) measured experiential acceptance, the extent to which one can accept unpleasant thoughts and emotions, 3) the

Cognitive Fusion Questionnaire (CFQ) (19) captured cognitive defusion, one's ability to view thoughts as transient phenomena.

Data were analyzed using SPSS version 22 (20). A median-split by LS was used to define high and low LS groups (split at SWLS = 17). *t*-tests/Mann-Whitney U tests were used to evaluate group differences.

Results

Sixteen mDMD participated. Average LS was in the "slightly below average" range (21). Average mood was in the minimal-to-mild range for anxiety (14) and depression (15). However, there was variation in all dependent variable scores (Table 1).

On average, mDMD view their illness as severe with extreme consequences, many symptoms, "permanence", and little treatment or personal control. Despite their accurate perception of the severity of DMD, their emotional response and level of concern were low, compared to asthma and diabetes reference groups (Supplementary Table S1, available online). Indeed, there was little difference between high and low LS groups in illness perceptions. Of 8 comparisons, only 1 showed a significant group difference (emotional representation; Table 1).

The Psychological Flexibility Model better differentiated high and low LS groups (Table 1). Those with higher LS, demonstrated greater ability to: accept unpleasant thoughts and feelings (experiential acceptance); step back from thoughts (cognitive defusion); and enable personally meaningful activity (valued-living). Here, between-group differences showed large

effect sizes. Anxiety and depression were worse in the low LS group. Advancing age, from 18 to 43 years, was not associated with LS (r = -0.10, P = 0.72), anxiety (r = -0.20, P = 0.45), or depression (r = 0.17, P = 0.52).

Discussion

This preliminary investigation supports the Psychological Flexibility Model. Thus, commensurate with previous qualitative studies (3, 8), the perceived ability to engage in personally meaningful activity, even with all that DMD brings, appears to best differentiate those with high LS. In addition, our results suggest that a cognitive style of accepting difficult thoughts and feelings (experiential acceptance) and stepping back from thoughts (cognitive defusion) characterized those with high LS. Such a cognitive style may help facilitate meaningful activity, especially in the context of DMD (7). Interestingly, despite having much greater functional impairment and reduced life expectancy, the average levels of concern and distress attributed to DMD (illness perceptions) were comparable to those of people with diabetes and asthma (Supplementary Table S1, available online). Therefore, in contrast to the Leventhal Self-Regulatory Model, which assumes that people with chronic disease continuously strive to make sense of their illness, it may be the case that mDMD are not particularly focused on their condition.

The clinical implications are that psychological interventions for those experiencing low LS, may be most effective if they enable personally meaningful activity, while facilitating accepting and open cognitive styles (7). However, larger, clinical samples and intervention studies are needed to confirm this finding.

This study has several limitations: 1) the small sample size meant only large effect sizes could be uncovered; 2) the online study and reliance on self-assessed inclusion and exclusion criteria meant that, although prohibited/unlikely, it is possible that, for example, participants with psychiatric disorders could have participated; 3) the use of a median split to create 2 groups; 4) unclear representativeness of the sample, for example, our group may have had higher than average IQ for mDMD (22).

Abbreviations

AAQ = Acceptance and Action Questionnaire

B-IPQ = Brief – Illness Perception Questionnaire

CFQ = Cognitive Fusion Questionnaire

ELS = Engaged Living scale

GAD-7 = Generalised Anxiety Disorder Questionnaire (7-item scale)

HAQ-DI = Health Assessment Questionnaire – Disability Index

LS = life satisfaction

mDMD = men with Duchenne muscular dystrophy

PHQ-9 = Patient Health Questionnaire (9-item scale)

SWLS = Satisfaction With Life Scale

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Table 1. Descriptive data relating to all measured variables, alongside details of sub-group analyses.

	Overall average (SD) N = 16	High Life satisfaction group average (SD) N= 8	High life satisfaction Observed range	Low Life satisfaction group average (SD) N= 8	Low life satisfaction Observed range	[a,b]*P*-value comparison between groups (effect size [*d*])
Age (years)	29.38 (6.61)	30.00 (6.33)	18 - 39	28.75 (7.27)	19 - 43	0.719 (0.20)
Functional impairment (HAQ-DI)[c]	3.00 (0.00)	3.00 (0.00)	3 -3	3.00 (3.00)	3 - 3	-
Life Satisfaction (SWLS)	18.00 (9.08)	26.00 (4.07)	18 - 30	10.00 (3.70)	5 - 16	>0.001 (4.40)
Anxiety (GAD-7)	4.13 (4.30)	1.63 (2.20)	0 - 5	6.63 (4.53)	3 - 13	0.018 (1.76)
Depression (PHQ-9)	4.88 (4.49)	2.38 (2.83)	0 - 6	7.38 (4.57)	3 - 14	0.022 (1.41)
Illness Perceptions (Brief IPQ)						
Consequences (*DMD has lots of consequences*)	9.38 (1.03)	9.13 (1.25)	7 - 10	9.63 (0.74)	8 - 10	0.574 (0.40)
Identity (*DMD has many symptoms*)	7.25 (2.46)	7.00 (1.69)	5 - 10	7.50 (3.16)	2 - 10	0.701 (0.24)
Timeline (*DMD will last forever*)	9.38 (1.71)	10.00 (0.00)	10 - 10	8.75 (2.32)	5 - 10	0.43 (0.76)
Personal Control (*I can control DMD*)	3.56 (3.03)	5.00 (2.45)	2 - 9	2.13 (3.00)	0 - 8	0.054 (1.12)
Treatment Control (*Treatment can control DMD*)	4.69 (3.67)	4.75 (3.81)	0 – 10	4.63 (3.78)	0 - 10	0.948 (0.04)
Coherence (*I understand my DMD*)	9.44 (1.37)	10.00 (0.00)	10 - 10	8.88 (1.81)	5 - 10	0.122 (1.33)
Concern (*DMD causes me much concern*)	5.25 (3.40)	4.50 (2.67)	0 - 8	6.00 (4.03)	0 - 10	0.396 (0.47)
Emotional Representation (*DMD causes me distress*)	4.75 (2.70)	3.13 (1.73)	1 - 5	6.38 (2.56)	2 - 10	0.021 (1.43)
Psychological Flexibility						
Values-based living (ELS) (*I know what's important and find ways to do this*)	51.31 (19.64)	65.25 (8.81)	49 - 73	40.85 (14.68)	20 - 59	0.001 (2.15)
Experiential Avoidance (AAQ) (*I avoid difficult thoughts and feelings – no matter the cost*)	34.38 (7.61)	29.63 (4.27)	22 - 35	39.13 (7.38)	27 - 51	0.007 (1.68)
Cognitive Fusion (CFQ) (*I am [stuck in] my thoughts*)	19.25 (6.99)	15.88 (5.49)	8 - 24	22.63 (6.97)	10 - 34	0.049 (1.15)

[a]where necessary, *t*-test *P*-values and effect sizes were adjusted for violation of homogeneity of variance assumption.
[b]where normality assumption violated, Mann-Whitney U tests were used instead of *t*-tests.
[c]Health Assessment Questionnaire – Disability Index (23)

Supplementary table S1. Reference values (possible ranges and scores in other populations) for included measures, available online

Measure	Possible range for the measure	Reference values from other populations: averages (SD)
Age (years)	18+	-
Functional impairment (HAQ-DI)[c]	0 - 3	Adult onset muscle disorders (24) = 1.51 (0.76)
Life Satisfaction (SWLS)	0 - 35	Undergraduate sample (13) = 23.5 (6.43)
Anxiety (GAD-7)	0 - 21	Patients without anxiety disorder (14) = 4.9 (4.8)
Depression (PHQ-9)	0 - 27	Patients without depressive disorder(15) = 3.3 (3.8)
Consequences (*DMD has lots of consequences*)	0 - 10	Asthma (16) = 3.5 (2.3) Diabetes (16) = 4.7 (2.9)
Identity (*DMD has many symptoms*)	0 - 10	Asthma (16) = 4.5 (2.3) Diabetes (16) = 4.6 (2.8)
Timeline (*DMD will last forever*)	0 - 10	Asthma (16) = 8.8 (2.2) Diabetes (16) = 9.2 (1.9)
Personal Control (*I can control DMD*)	0 - 10	Asthma (16) = 6.7 (2.4) Diabetes (16) = 6.7 (2.3)
Treatment Control (*Treatment can control DMD*)	0 - 10	Asthma (16) = 7.9 (2.0) Diabetes (16) = 8.0 (2.3)
Coherence (*I understand my DMD*)	0 - 10	Asthma (16) = 6.5 (2.6) Diabetes (16) = 7.9 (2.3)
Concern (*DMD causes me much concern*)	0 - 10	Asthma (16) = 4.6 (2.8) Diabetes (16) = 7.0 (3.1)
Emotional Representation (*DMD causes me distress*)	0 - 10	Asthma (16) = 3.3 (2.9) Diabetes (16) = 4.3 (3.3)
Values-based living (ELS) (*I know what's important and find ways to do this*)	16 - 80	Community sample (17) = 60.80 (7.83)
Experiential Avoidance (AAQ) (*I avoid difficult thoughts and feelings – no matter the cost*)	7 - 63	Male university students (18) = 32.7 (6.4)
Cognitive Fusion (CFQ) (*I am [stuck in] my thoughts*)	7 - 49	Community sample (19) = 22.28 (8.30)