

Psychosocial Factors and Well-Being Among Adults With Charcot–Marie–Tooth Disease: A Cross-Sectional Study

Payton D. Rule and Patrick L. Hill

Department of Psychological and Brain Sciences, Washington University in St. Louis

Purpose/Objective: The goal of the current study is to examine the relationships between psychosocial factors and well-being among adults with Charcot–Marie–Tooth (CMT) disease, a progressive neuromuscular disorder. Specifically, we aimed to examine associations between psychosocial factors and sense of purpose as well as the moderating role of sense of purpose on associations between psychosocial factors and two other measures of well-being (life satisfaction and health-related quality of life). **Research Method/Design:** In 2021, we recruited 263 U.S. adults with CMT ($M_{\text{age}} = 60.15$ years, 58.56% female, 92.40% White) to take part in a cross-sectional study. Participants were asked to complete an online survey assessing components of well-being as well as various psychosocial factors. **Results:** Moderate-to-strong associations were found between most psychosocial factors assessed and sense of purpose in individuals with CMT. In addition, sense of purpose moderated the relationship between multiple psychosocial factors and other measures of well-being. **Conclusions/Implications:** Psychosocial factors may be important to consider when examining well-being among individuals with CMT. Furthermore, sense of purpose may be a beneficial tool for promoting well-being in this population.

Impact and Implications

This study adds to the literature by investigating well-being among adults with Charcot–Marie–Tooth (CMT) disease, a population that has been underrepresented in psychology research. The results of this study could help inform adaptations to existing programs and resources to help promote well-being among individuals with CMT disease. CMT is a hereditary peripheral neuropathy affecting roughly one in 2,500 people worldwide (Charcot–Marie–Tooth Association, 2021). CMT affects the peripheral nervous system and leads to progressive muscle weakness, decreased balance, fatigue, pain, and gradual loss of physical functioning (Taniguchi et al., 2013). Past research on well-being in people with CMT is very limited; however, studies suggest that people with CMT may have worse mental health and well-being than those without CMT. Multiple studies have found that individuals with CMT experience decreased quality of life in physical, psychological, and social domains compared to the general population (Burns et al., 2010; Taniguchi et al., 2013; Vinci et al., 2005). In addition, those with CMT may have a higher risk for developing depressive symptoms than those without CMT, although studies have shown mixed results (Cordeiro et al., 2014). Researchers believe that these adverse effects on mental health and well-being are secondary to CMT and are not caused by the biological mechanisms of the disorder itself (Shy & Rose, 2005). Instead, decreases in well-being likely stem from psychological, social, and physical factors or difficulties that arise because of CMT symptoms. These could include anxiety surrounding the progressive nature of the disorder, social stigmatization, or experiences of disability discrimination. While past work has evaluated well-being in adults with CMT, we are unaware of any studies that have examined the relationship between specific psychosocial factors and well-being in this population. The current study will address this by investigating the associations between psychosocial factors and sense of purpose as well as the moderating role of sense of purpose on associations between psychosocial factors and two other measures of well-being (life satisfaction and health-related quality of life).

Payton D. Rule identifies as disabled and holds a volunteer role with the Charcot–Marie–Tooth Association. The authors have no other conflicts of interest to disclose. Recruitment for this study was completed in collaboration with the Charcot–Marie–Tooth Association. This study’s design, hypotheses, and analyses were preregistered (see https://osf.io/hc384/?view_only=c6dfeca870e3402cbee9cb4121f14965). Materials and analysis code for this study are available by emailing the corresponding author. Supplemental materials can be viewed at https://osf.io/z562a/?view_only=1f63a856cc254dff8077c60ec0cb38af.

Payton D. Rule served as lead for conceptualization, data curation,

formal analysis, investigation, methodology, project administration, resources, validation, visualization, writing—original draft, and writing—review and editing. Patrick L. Hill served as lead for supervision and served in a supporting role for conceptualization, data curation, formal analysis, investigation, methodology, project administration, resources, validation, visualization, writing—original draft, and writing—review and editing.

Correspondence concerning this article should be addressed to Payton D. Rule, Department of Psychological and Brain Sciences, Washington University in St. Louis, 1 Brookings Dr. Campus Box 1125, St. Louis, MO 63130, United States. Email: rulep@wustl.edu

Keywords: disabilities, well-being, psychosocial factors

Factors That Promote Well-Being

Health psychology research has identified a variety of psychosocial factors that are associated with well-being in people with chronic health conditions. Specifically, studies have found that social support, friendships with other people with disabilities, adaptability, and self-efficacy are all linked to higher well-being in clinical populations (Börsbo et al., 2010; Silverman et al., 2017). In contrast, experiencing disability discrimination and higher levels of anxiety have been negatively associated with well-being (Hackett et al., 2020; Hsu et al., 2019). As such, psychosocial factors may be critical to consider when examining well-being among individuals with Charcot–Marie–Tooth (CMT) disease.

One underexamined component of well-being that may be especially beneficial for people with CMT is sense of purpose. McKnight and Kashdan (2009) describe having a purpose as an overarching life aim that motivates and directs ongoing goal setting, pursuit, and attainment, and which can result in an increased sense of meaning. Multiple researchers suggest that purpose often becomes intertwined with an individual's identity and personality, influencing many different areas of their life (Hill & Burrow, 2012; McKnight & Kashdan, 2009). Most empirical work has focused on whether one feels a sense of purpose, which can be described as the degree to which a person feels an overarching sense of direction and is engaged in valued activities (Ryff, 1989; Scheier et al., 2006). The pursuit of valued activities provides a sense of purpose and a means of staying behaviorally engaged with life, which is supported by work connecting sense of purpose with objective physical activity (Hooker & Masters, 2016).

Purpose and Physical Health

Prior research has suggested that sense of purpose is positively associated with a variety of physical health measures. Participants with a higher sense of purpose reported better self-rated health, less physical limitations, and less role disruptions due to their physical health (Scheier et al., 2006). Moreover, this relationship between sense of purpose and self-rated health was also present in a clinical population, individuals with diabetes, and was not moderated by diabetes status (Weston et al., 2019). This finding suggests that even individuals living with a chronic disease, disability, or health condition can benefit from sense of purpose. In addition to self-rated health, sense of purpose has been associated with more objective measures of physical health such as walking speed and grip strength (Kim et al., 2017). Along with better self-rated health and less physical limitations, sense of purpose has also been linked to a reduced risk of early cardiovascular disease and mortality risk in younger and older adults (Cohen et al., 2016; Hill & Turiano, 2014; Kim et al., 2019). This association with longevity is present even when accounting for chronic medical conditions, depressive symptoms, and other risk factors (Boyle et al., 2009). One potential explanation for the relationship between sense of purpose and physical health may be engagement in health-promoting behaviors. Prior research has suggested that a higher sense of purpose is associated with more physical activity, better nutrition, better sleep quality, and regular use of preventative health care, all of which can positively impact health outcomes (Hill et al., 2019; Kim, Chen, et al., 2022; Kim et al., 2014, 2019).

Purpose and Mental Health

Prior research has suggested that sense of purpose may promote better mental health. One study found that sense of purpose increased mindfulness and behavioral activation among participants and led to less depressive and anxious symptoms (Crego et al., 2021). Furthermore, research suggests that sense of purpose may help promote resilience and recovery among those who have experienced severe, often repeated trauma (Alim et al., 2008). Additionally, studies have shown that individuals with a higher sense of purpose may be better able to cope with daily stress and tend to perceive obstacles as less difficult than those with a lower sense of purpose (Burrow et al., 2016; Hill et al., 2018). Relatedly, prior research has found that those with a higher sense of purpose experience more hope, life satisfaction, and social support (Bronk et al., 2009; Heisel & Flett, 2004; Ryff & Keyes, 1995; Ulmer et al., 1991).

Purpose and Adversity

Previous work has shown that sense of purpose may offer an important pathway for coping with and overcoming obstacles by increasing prospection about the future (Bronk & Mitchell, 2022). Because sense of purpose includes a sense of direction and life engagement, it requires that individuals look to their future to plan activities that align with their values and sense of direction. In a study examining the relationship between future time perspective and sense of purpose, researchers found that those who perceived their future time as more limited had a lower sense of purpose (Pfund et al., 2022). Imagining one's future path and planning how to reach goals may provide a sense of meaning and help cultivate perseverance and grit. Research also found that individuals who have a stronger sense of purpose report higher levels of grit (Hill et al., 2016). By maintaining a more long-term and future-oriented perspective, individuals with a higher sense of purpose may feel more motivated and equipped to weather life's storms and persevere when encountering obstacles.

Although somewhat limited, past research suggests that greater sense of purpose may lead to better adjustment and quality of life in people with chronic illnesses and disabilities. One study found that those with a higher sense of purpose experienced better adjustment after a spinal cord injury (Thompson et al., 2003). Additional studies have found that sense of purpose and meaning in life may be a more important predictor of well-being than one's limitations or symptoms. For instance, in a study of well-being in people who had sustained a spinal cord injury, individuals who perceived greater functional loss reported lower well-being (deRoon-Cassini et al., 2009). However, this negative association weakened significantly for those with a greater sense of purpose and meaning in life. This provides further evidence of the importance of psychosocial factors for determining well-being in those with health conditions. Similar findings have also been reported in other health-related populations. In one study of quality of life among individuals with rheumatoid arthritis, researchers found that higher sense of purpose did not predict better physical health but did predict better mental health quality of life (Verduin et al., 2008). This provides further evidence that sense of purpose may be a powerful tool for promoting better coping and well-being among individuals with physical health conditions and disabilities.

Current Study

Considering that CMT may lead to worse well-being (Burns et al., 2010; Cordeiro et al., 2014; Taniguchi et al., 2013; Vinci et al., 2005), the current study examined connections between psychosocial factors and well-being in people with CMT. Specifically, the first goal of this study was to examine the relationships between psychosocial factors and sense of purpose among people with CMT. Nine psychosocial factors were examined: discrimination, CMT-related anxiety, avoidance due to CMT-related anxiety, self-efficacy, adaptive device use, adaptive device helpfulness, CMT/disability community perceived support, involvement in the CMT/disability community, and perceived social support. The second goal of this study was to examine any moderating effects of sense of purpose on the relationships between psychosocial factors and two measures of well-being, life satisfaction and health-related quality of life (HRQoL), in people with CMT.

We predicted that a relationship would exist between sense of purpose and each psychosocial factor. Specifically, we hypothesized that sense of purpose would be positively correlated with self-efficacy, adaptive device use, adaptive device helpfulness, CMT/disability community perceived support, involvement in the CMT/disability community, and perceived social support. In contrast, we predicted that sense of purpose would be negatively correlated with CMT-related anxiety, avoidance due to CMT-related anxiety, and discrimination. Additionally, exploratory analyses examined sense of purpose as a moderator of the relationship between two measures of well-being (life satisfaction and HRQoL) and each psychosocial factor. We predicted that people with a higher sense of purpose would experience stronger positive associations and weaker negative associations between psychosocial factors and measures of well-being.

Method

Participants

Participants included 315 adults with CMT ($M_{\text{age}} = 59.26$, 59.27% female, 91.84% White) who took part in an approximately 10- to 20-min online survey. Some participants failed to meet eligibility criteria (<18 years of age, no CMT diagnosis, or lived outside of the United States; $n = 20$) or did not complete any of the study measures ($n = 32$) and were excluded from the analytic sample. Our final sample included 263 U.S. adults with CMT ($M_{\text{age}} = 60.15$ years, 58.56% female, 92.40% White) who completed at least one study measure. Twenty participants failed to complete all measures and thus were only included in some analyses. More information regarding sample demographics is shown in Table 1.

Measures

Sense of Purpose

Sense of purpose was assessed using the six-item Life Engagement Test (Scheier et al., 2006). Participants were asked to respond with their agreement with each statement on a scale of 1 (*strongly disagree*) to 5 (*strongly agree*). An example item is "To me, the things I do are all worthwhile." These six items were averaged to create a sense of purpose score with higher scores indicating a higher sense of purpose ($\alpha = .85$). Composite means and standard

Table 1
Sample Demographic Characteristics

Characteristics	<i>n</i>	Percentage
Race		
White	243	92.40
Hispanic or Latinx	4	1.52
Asian	4	1.52
Biracial/multiracial	7	2.66
Other	5	1.90
Gender		
Female	154	58.56
Male	107	40.68
Genderqueer	1	0.38
Prefer not to answer	1	0.38
Income		
Under \$10,000	2	0.82
\$10,000–\$24,999	23	9.47
\$25,000–\$49,999	23	9.47
\$50,000–\$74,999	67	27.57
\$75,000–\$99,999	37	15.23
\$100,000–\$124,999	30	12.35
\$125,000–\$150,000	16	6.58
Over \$150,000	45	18.52
	<i>M</i>	<i>SD</i>
Age (in years)	60.15	15.75
Time since diagnosis (in years)	24.57	17.82
Age at symptom onset (in years)	23.87	20.15
Physical functioning (subscale of SF-36)	40.66	26.85

Note. SF-36 = Official Short Form-36.

deviations for all variables are shown in Table 2. In the original validation paper, this scale exhibited reliability and validity across samples with health conditions.

Life Satisfaction

Life satisfaction was assessed using the five-item Satisfaction with Life Scale (Diener et al., 1985). Participants were asked to respond with their agreement with each statement on a scale of 1 (*strongly disagree*) to 7 (*strongly agree*). An example item is, "In most ways my life is close to my ideal." These five items were averaged to create a life satisfaction score with higher scores indicating higher life satisfaction ($\alpha = .88$). This scale has shown high internal consistency and reliability across different samples, including samples with health conditions (Pavot & Diener, 2009).

HRQoL

HRQoL was assessed using the Official Short Form-36 (SF-36) measure (Ware & Sherbourne, 1992; developed at RAND as part of the Medical Outcomes Study). Composite scores were calculated for each of the eight health concepts captured in this measure: physical functioning, bodily pain, role limitations due to physical health problems, role limitations due to personal or emotional problems, emotional well-being, social functioning, energy/fatigue, and general health perceptions. As shown in Table 2, all subscales were correlated to purpose similarly. Therefore, for moderation analyses, an overall HRQoL score was calculated by averaging scores from the eight subscales to reduce the number of statistical tests. Higher scores indicate higher HRQoL ($\alpha = .84$). Use of an overall HRQoL score has

This document is copyrighted by the American Psychological Association or one of its allied publishers. This article is intended solely for the personal use of the individual user and is not to be disseminated broadly.

Table 2
Correlation Table and Mean and SD Scores for Psychosocial and Well-Being Variables

Variable	Sense of purpose	Life satisfaction	HRQoL	Discrimination	CMT-related anxiety	Perceived social support	CMT/disability community perceived support	Avoidance due to CMT-related anxiety	Involvement in the CMT or disability communities	Adaptive device helpfulness	Adaptive device use	Self-efficacy
Sense of purpose	1											
Life satisfaction	0.55** (257)	1										
HRQoL	0.51** (257)	0.62** (260)	1									
Discrimination	-0.26** (255)	-0.26** (255)	-0.33** (255)	1								
CMT-related anxiety	-0.32** (245)	-0.33** (245)	-0.49** (245)	0.30** (245)	1							
Perceived social support	0.43** (251)	0.47** (251)	0.32** (251)	-0.26** (251)	-0.16* (245)	1						
CMT/disability community perceived support	0.19** (244)	0.26** (244)	0.09 (244)	-0.18** (244)	0.01 (243)	0.44** (244)	1					
Avoidance due to CMT-related anxiety	-0.33** (246)	-0.38** (246)	-0.49** (246)	0.25** (246)	0.54** (245)	-0.21** (246)	-0.08 (244)	1				
Involvement in the CMT or disability communities	0.01 (246)	-0.02 (246)	-0.05 (246)	0.00 (246)	0.05 (245)	-0.03 (246)	0.27** (244)	0.01 (246)	1			
Adaptive device helpfulness	0.17 (188)	0.10 (188)	0.05 (188)	-0.26** (188)	0.04 (188)	0.17* (188)	0.31** (188)	-0.05 (188)	0.02 (188)	1		
Adaptive device use	-0.15* (244)	-0.16** (244)	-0.32** (244)	0.10 (244)	0.23** (243)	-0.07 (244)	0.25** (244)	0.22** (244)	0.17** (244)	-0.18** (244)	1	
Self-efficacy	0.47** (249)	0.55** (249)	0.54** (249)	-0.31** (249)	-0.53** (245)	0.38** (249)	0.22** (244)	-0.49** (246)	-0.05 (246)	0.13 (188)	-0.18** (244)	1
Physical functioning	0.24** (256)	0.37** (259)	0.63** (260)	-0.08 (254)	-0.42** (244)	0.11 (250)	-0.04 (243)	-0.38** (245)	-0.06 (245)	-0.12 (187)	-0.46** (243)	0.32** (248)
Role limitation due to physical health problems (higher scores = less limitation)	0.24** (256)	0.37** (259)	0.73** (260)	-0.17** (254)	-0.29** (245)	0.17** (250)	0.01 (244)	-0.35** (246)	-0.06 (246)	-0.04 (188)	-0.25** (244)	0.35** (248)
Role limitation due to personal or emotional problems (higher scores = less limitation)	0.39** (255)	0.39** (258)	0.73** (259)	-0.25** (253)	-0.34** (244)	0.22** (250)	0.04 (243)	-0.27** (245)	-0.01 (245)	-0.06 (187)	-0.16** (243)	0.29** (248)
Energy/fatigue (higher scores = less fatigue)	0.39** (257)	0.53** (260)	0.76** (261)	-0.21** (255)	-0.35** (245)	0.28** (251)	0.09 (244)	-0.35** (246)	-0.06 (246)	0.04 (188)	-0.17** (244)	0.45** (249)
Emotional well-being	0.55** (257)	0.52** (260)	0.59** (261)	-0.33** (255)	-0.34** (245)	0.34** (251)	0.10 (244)	-0.29** (246)	0.03 (246)	0.17** (188)	-0.03 (244)	0.44** (249)
Social functioning	0.52** (257)	0.58** (260)	0.83** (261)	-0.34** (255)	-0.44** (245)	0.37** (251)	0.14 (244)	-0.49** (246)	-0.11 (246)	0.03 (188)	-0.29** (244)	0.54** (249)
Bodily pain (higher scores = less pain)	0.30** (257)	0.36** (260)	0.65** (261)	-0.26** (255)	-0.30** (245)	0.25** (251)	0.18** (244)	-0.35** (246)	0.03 (246)	0.11 (188)	-0.24** (244)	0.34** (249)
General health	0.36** (257)	0.53** (260)	0.70** (261)	-0.27** (255)	-0.34** (245)	0.16* (251)	0.04 (244)	-0.33** (246)	0.01 (246)	0.07 (188)	-0.20** (244)	0.42** (249)
Time since diagnosis (years)	-0.04 (253)	0.03 (256)	-0.04 (257)	-0.03 (251)	0.11 (241)	-0.03 (247)	0.16** (240)	0.15* (242)	0.002 (242)	-0.01 (188)	0.18** (240)	0.05 (245)
Time since symptom onset (years)	-0.08 (253)	-0.10 (256)	-0.13* (257)	0.07 (251)	0.12* (241)	-0.04 (247)	0.14* (240)	0.15** (242)	0.05 (242)	0.09 (188)	0.20** (240)	-0.05 (245)
M	3.91	4.43	52.96	1.75	3.09	5.51	3.52	3.18	1.60	4.23	1.22	2.95
SD	0.72	1.44	19.20	0.78	0.86	1.25	0.72	0.97	0.49	0.91	0.42	0.90

Note. Degrees of freedom for each analysis are shown in parentheses. HRQoL = health-related quality of life; CMT = Charcot-Marie-Tooth disease. * $p < .05$. ** $p < .01$.

been reported frequently in the literature, with the average of eight subscales being the most frequent strategy for combining these measures (Lins & Carvalho, 2016). All subscales have demonstrated high validity and reliability and have been used extensively in clinical populations (Bousquet et al., 1994; Lyons et al., 1994; Moorer et al., 2001; Pinar, 2005; Ware et al., 1994).

Perceived Social Support

Perceived social support was assessed using the 12-item Multidimensional Scale of Perceived Social Support (Zimet et al., 1988). Participants were asked to respond with their agreement with each statement on a scale of 1 (*very strongly disagree*) to 7 (*very strongly agree*). An example item is, "There is a special person who is around when I am in need." Items were averaged to create a perceived social support score with higher scores indicating higher perceived social support ($\alpha = .95$). This scale has been used extensively and has demonstrated good reliability and validity in clinical populations across a variety of health conditions (Kim, Yeom, & Jung, 2022; Nearchou et al., 2022).

Self-Efficacy

Self-efficacy was assessed using the six-item University of Washington Self-Efficacy Scale (Amtmann et al., 2012) adapted for the CMT population. Participants were asked to respond with their confidence regarding each statement on a scale of 1 (*not at all*) to 5 (*completely*). An example item is, "You can keep your CMT from interfering with your ability to deal with unexpected events." These six items were averaged to create a self-efficacy score with higher scores indicating higher self-efficacy ($\alpha = .89$). This scale is one of the most widely utilized self-efficacy measures in the health literature and has exhibited good reliability and validity across samples with a variety of health conditions (Amtmann et al., 2012; Chung et al., 2016).

Discrimination

Experiences of discrimination were assessed using the Everyday Discrimination Scale (Williams et al., 1997). This scale contains nine items that ask participants to respond with how often they experience different discrimination-related events on a scale of 1 (*almost every day*) to 6 (*never*). An example item is, "You are treated with less courtesy than other people are." After reverse scoring, these nine items were averaged to create a discrimination score with higher scores indicating more experiences of discrimination ($\alpha = .89$). A final item in this scale asked participants to select all factors they attributed to their experiences of discrimination (e.g., their weight, gender, CMT, etc.). This scale has been validated and used extensively in population health research (Krieger et al., 2005; Williams & Mohammed, 2009). In addition, this measure is associated with constructs, such as depression, that research has found to be closely related to discrimination (Britt-Spells et al., 2018; Lee et al., 2022).

CMT-Related Anxiety

Participants were asked how much anxiety they experience about six CMT-related things on a scale of 1 (*none*) to 5 (*a lot*). An example item is "How much anxiety do you experience about tripping/falling due to your CMT?" These six items were averaged to create a CMT-related anxiety score. Higher scores indicate higher levels of

CMT-related anxiety ($\alpha = .80$). Additional questions from this measure were used to examine two other factors: avoidance due to CMT-related anxiety and ranked anxiety. Avoidance was assessed by asking participants "How often do you avoid situations or activities due to anxiety related to your CMT symptoms?" on a scale of 1 (*never*) to 5 (*a lot*). Lastly, participants were asked to rank the six items listed from most to least anxiety-inducing. These scales were created for the current study (see Appendix A for measure items).

CMT/Disability Community Involvement

Involvement was assessed by asking participants, "Within the past two years, have you been involved with the CMT or broader disability communities?" In addition, participants who responded "yes" were then asked to list the activities they have been involved in and the length of time they have been involved.

CMT/Disability Community Perceived Support

The support participants perceived from the CMT and disability communities was measured by asking participants to respond with their agreement to five statements regarding their connection/belonging on a scale of 1 (*strongly disagree*) to 5 (*strongly agree*). An example item is "I feel a sense of belonging within the CMT community." These six items were averaged to create a CMT/disability community perceived support score. This scale was created for the current study (see Appendix B for measure items). Higher scores indicate higher perceived support ($\alpha = .75$).

Adaptive Device Use

This measure was used to report two scores regarding adaptive devices: use and perceived helpfulness. Adaptive device use was assessed by asking participants "Do you currently use any adaptive devices?" Adaptive device helpfulness was measured by asking participants to rate how helpful they find their adaptive devices on a scale of 1 (*not at all helpful*) to 5 (*extremely helpful*). These items were created for the current study.

Procedures

Participants were recruited in 2021 via social media posts made by the Charcot-Marie-Tooth Association (CMTA) on their Twitter, Instagram, and Facebook Group pages and email blasts to the CMTA's database. Once they clicked on the survey link, participants were directed to a consent information sheet. To confirm eligibility, once they consented to participation, participants were asked to self-report whether they had a diagnosis of CMT, were at least 18 years of age, and resided in the United States. Participants then completed the survey which contained the measures described above. No compensation was provided to participants; however, upon completion of the survey, participants were given the option to be entered into a raffle for one of two \$50 gift cards. The sample displayed no evidence of straight-line responding (i.e., providing the same response for each item) or completing the survey in under a minute, solely to receive credit.

Analytic Plan

Statistical Models. To test hypotheses regarding the relationships between psychosocial variables and sense of purpose,

correlation analyses were conducted between each psychosocial variable and sense of purpose. To receive a composite score for a particular measure, participants must have responded to at least 75% of the items in that measure. However, it is worth noting that the vast majority of participants (~90%) responded to all measure items. An alpha of .05 was used throughout the analyses.

Exploratory Analyses. Moderation analyses were conducted to examine whether sense of purpose is a moderator between psychosocial variables and two other measures of well-being (life satisfaction and HRQoL). In addition, partial correlations for all variables were calculated when controlling for age, income, age at diagnosis, and age at symptom onset to test whether associations were held. Exploratory correlations were calculated between time since diagnosis and sense of purpose as well as time since symptom onset and sense of purpose.

Transparency and Openness

All analyses were conducted in R utilizing the psych and ppcor packages. This research was considered exempt by the Washington University in St. Louis Institutional Review Board due to the anonymity of participants and lack of experimental manipulation. This study design, hypotheses, and analyses were preregistered; see <https://doi.org/10.17605/OSF.IO/HC384>. Materials and analysis code for this study are available by emailing the corresponding author. Supplemental materials can be viewed at https://osf.io/z562a/?view_only=fd022b2bb57d4828b3d1c55c83014cff.

Results

Descriptive Statistics and Correlations

As shown in Table 2, participants reported moderate-to-high levels of CMT-related anxiety ($M = 3.09$, $SD = 0.86$) and avoidance due to their CMT-related anxiety ($M = 3.18$, $SD = 0.97$). Almost half of the participants (47.95%) reported experiencing the most anxiety about the progressive nature of their CMT. In addition, over a third of our sample (37.40%) reported experiencing discrimination due to their CMT. Nevertheless, on average, people with CMT reported experiencing a high sense of purpose, with most participants scoring above the scale midpoint ($M = 3.91$, $SD = 0.72$).

As shown in Table 2, medium to very large correlations were observed between a variety of psychosocial variables and sense of purpose.¹ Of the psychosocial variables, self-efficacy had the strongest association with sense of purpose. Contrary to our hypothesis, no correlation was found between involvement in the CMT/disability communities and sense of purpose. Additionally, adaptive device use was negatively correlated with sense of purpose.

Exploratory Analyses

Age Since Diagnosis and Onset

As seen in Table 2, correlations were calculated between sense of purpose and time since diagnosis as well as sense of purpose and time since symptom onset. Neither time since symptom onset nor time since diagnosis was significantly correlated with sense of purpose, suggesting that these variables may not have a large influence on one's sense of purpose.

Partial Correlations

Partial correlations were conducted to control for the effects of age, income, age at diagnosis, and age at symptom onset on the relationships between psychosocial variables and sense of purpose. All correlations remained significant when controlling for these demographics, suggesting that these factors do not play a large role in these associations. Presentation of these analyses can be found at https://osf.io/z562a/?view_only=fd022b2bb57d4828b3d1c55c83014cff.

Moderation Analyses

Figure 1 shows the results of moderation analyses to explore the moderating role of sense of purpose in the relationships between psychosocial variables and well-being (life satisfaction and HRQoL). First, as shown in Figure 1A, perceived social support was more strongly associated with greater life satisfaction for those with a higher sense of purpose. As shown in Figure 1B, a moderation effect was also found for the relationship between CMT/disability community perceived support and HRQoL. CMT/disability community perceived support was more associated with greater HRQoL for those with a higher sense of purpose. Additionally, as shown in Figure 1C, sense of purpose moderated the relationship between self-efficacy and HRQoL in a similar pattern: for those with higher sense of purpose, self-efficacy was more positively associated with HRQoL. In addition, sense of purpose also appeared to moderate the relationship between CMT-related anxiety and HRQoL. As displayed in Figure 1D, those with high CMT-related anxiety reported lower HRQoL; however, this negative correlation was stronger for those with high sense of purpose. Because these were exploratory analyses, no familywise error correction was made for the models presented. Full presentation of these analyses is available at https://osf.io/z562a/?view_only=fd022b2bb57d4828b3d1c55c83014cff.

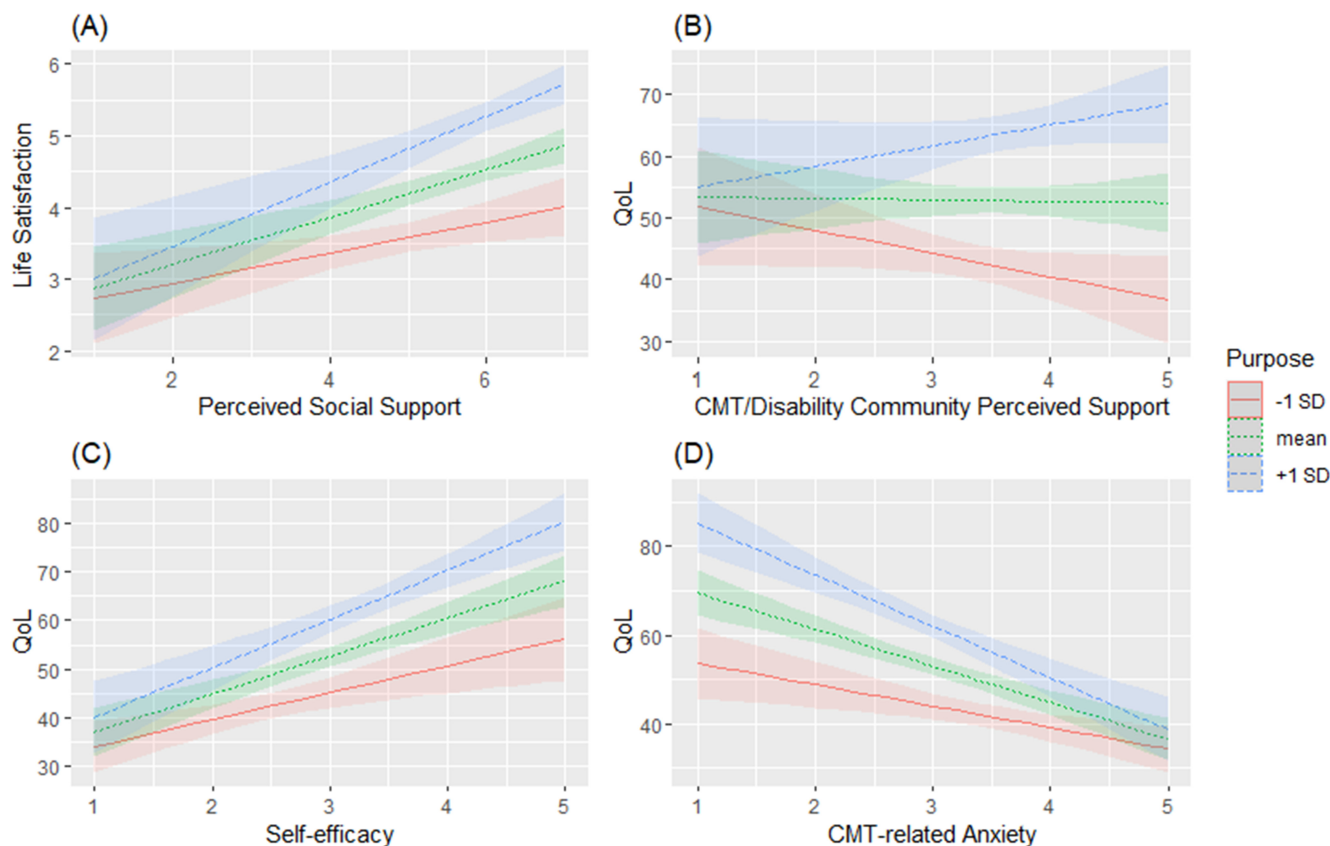
Discussion

This study was one of the first to examine sense of purpose in individuals with CMT. Specifically, this study explored the relationships between psychosocial factors and sense of purpose among adults with CMT as well as the impact of sense of purpose on associations between psychosocial factors and two other measures of well-being. The results suggested many psychosocial factors that are strongly associated with sense of purpose in people with CMT. Furthermore, this study found evidence of the moderating role of sense of purpose in the relationships between various psychosocial factors and well-being. Lastly, the measures utilized in the current study displayed evidence of good reliability and validity. Specifically, all composite measures showed good scale reliability, including the composite measures created for the current study. In addition, correlational analyses provide evidence of measurement validity, insofar that associations between constructs align with conceptual expectations. For instance, the measures created for the current study (e.g., community

¹ Correlations remained stable when excluding the 20 participants who did not receive composite scores on each measure. The largest difference in magnitude was a strengthening of a correlation by 0.04; there was no change in p -value cutoffs shown in Table 2.

Figure 1

Interaction Plots for the Associations Between Psychosocial Factors at Different Levels for Sense of Purpose (± 1 SD or at the M for the Sample)



Note. (A) perceived social support and life satisfaction; (B) CMT/disability community perceived support and HRQoL; (C) self-efficacy and HRQoL; (D) CMT-related anxiety and HRQoL. QoL = health-related quality of life; CMT = Charcot-Marie-Tooth disease. See the online article for the color version of this figure.

involvement and adaptive device use) were not associated with well-being indicators, demonstrating their distinction. Moreover, CMT-related anxiety was associated but not synonymous with broader indicators of well-being, such as emotional well-being, suggesting participants were reporting differentially across these scales. This provides evidence these measures can be applied to individuals with CMT. Full presentation of these analyses is available in Table 2. The following sections provide a further review of the findings of the study, their implications, and limitations.

Psychosocial Variables and Well-Being

In alignment with our hypotheses, the current study found associations between many psychosocial factors and sense of purpose. As predicted, self-efficacy, CMT/disability community perceived support, and perceived social support were positively correlated with sense of purpose. Likewise, CMT-related anxiety and discrimination were negatively correlated with sense of purpose. Contrary to our hypothesis, there was no association between involvement in the CMT/disability communities and sense of purpose. Since previous work has found social support to be a powerful predictor of psychological well-being, it was expected that involvement in the CMT or

disability communities could be another means of gaining support and promoting a sense of purpose (Ulmer et al., 1991). However, the lack of association between these variables suggests that this may not always be the case. There are at least two reasons why no relationship was found between involvement in the CMT/disability communities and sense of purpose. First, it is possible that involvement is helpful for certain people and harmful for others. For someone newly diagnosed with CMT, attending support group meetings with others who are more severely affected could lead to more worry or dread and negatively impact their sense of purpose. However, for someone feeling alone in their struggles, a support group could be a way of gaining support and resources that could increase their sense of purpose. Second, the subjective perception of support or length of involvement may be more important for well-being than whether they are involved. Differing types of support offered by various groups or organizations may have unique effects on one's sense of purpose. For example, a disability support group may focus on providing emotional support and resources while an adaptive sports team may focus on team bonding and athletic training. In addition, people who are involved in their group or organization longer may feel more connected and supported and thus might experience a greater sense of purpose. Future research is needed to further

examine individual differences in the associations between involvement and sense of purpose and whether sense of purpose varies based on the capacity or length of time in which an individual is involved.

In addition, counter to predictions, participants in the current study who reported using adaptive devices had a lower sense of purpose. This unexpected result may be due to the increased likelihood for those with more progressed or severe CMT to utilize adaptive devices. This aligns with prior research findings that individuals with more physical limitations often report lower well-being (Kim et al., 2017; Scheier et al., 2006). An alternative explanation is that many individuals utilizing adaptive devices may view them as a marker of limitation. In society, adaptive devices such as wheelchairs are often viewed as symbols of limitation rather than a means of mobility and freedom. If people hold negative views of their adaptive devices, utilizing them may cause embarrassment, sadness, or shame and negatively impact sense of purpose. Future research is needed to further investigate the association between adaptive device use and sense of purpose while controlling for CMT severity and perceptions of adaptive devices. In addition, future research should examine whether certain adaptive devices have a greater impact on sense of purpose and if this varies based on an individual's symptoms and severity.

Sense of Purpose as a Moderator

In our exploratory analyses, we investigated the moderating role of sense of purpose on the relationships between psychosocial factors and two measures of well-being, life satisfaction and HRQoL. Results indicated that sense of purpose moderated the relationship between CMT-related anxiety and HRQoL. While people with higher CMT-related anxiety reported a lower HRQoL, for those reporting CMT-anxiety symptoms up to 0.5 standard deviations above the mean, this association was reduced for people with a higher sense of purpose. Prior research has suggested that sense of purpose may help cultivate a future-oriented perspective and resilience, both of which may aid in better managing one's anxiety and lead to greater well-being (Bronk & Mitchell, 2022; Horowitz & Vanner, 2010). This finding has implications for future work on well-being interventions as it identifies sense of purpose as a potential target to promote better coping with anxiety and improve well-being in people with CMT.

Sense of purpose also was found to moderate the relationship between CMT/disability community perceived support and HRQoL. For people with a high sense of purpose, more perceived support was associated with a greater HRQoL. Interestingly, for participants with a low sense of purpose, higher perceived support was associated with lower HRQoL. This could reflect differences in how purposeful participants view their connections to the CMT community. People with a high sense of purpose may view their connections to the CMT community and support within the community as a valuable source of purpose, and they may utilize those connections as motivation to pursue personally valued goals or activities. For example, a purposeful individual who feels a connection to the CMT or disability communities may be motivated to engage in advocacy to bring awareness to ableism or discrimination experienced by others in the community. In contrast, people with a low sense of purpose may not derive motivation from experiences of discrimination or ableism and instead perceive them as unchangeable aspects of their membership to the CMT or disability communities. Therefore, an individual with a low

sense of purpose who feels a high sense of belonging and connection to the CMT or disability communities may experience worse well-being. This aligns with prior research findings suggesting that individuals with a higher sense of purpose have higher levels of grit and more motivation to engage in personally valued activities than those with a lower sense of purpose (Hill et al., 2016; McKnight & Kashdan, 2009). More research is needed to examine differences in the ways people with a higher sense of purpose perceive their connection, belonging, and support in the CMT and disability communities compared to those with a lower sense of purpose.

Program and Clinical Implications

This study is one of the first to examine the relationship between psychosocial factors and components of well-being in a population of adults with CMT. As such, it contributes valuable knowledge to the CMT, medical, and scientific communities to help inform changes to existing programs and clinical practices to promote better well-being in this population. First, CMT programs and resources could be adapted to help individuals better cope with detrimental psychosocial factors. Our results suggest that CMT-related anxiety and discrimination could lead to lower sense of purpose, yet our results also show that these are common experiences for people with CMT. While research is needed to determine beneficial interventions for CMT-related anxiety and discrimination, one promising therapy is behavioral activation. Behavioral activation focuses on helping individuals engage in pleasurable or personally valued activities (Dimidjian et al., 2011; Hooker et al., 2020). Prior studies in non-CMT samples have shown behavioral activation to be a promising treatment for generalized anxiety and to aid in better coping with experiences of discrimination (Boswell et al., 2017; Mekawi et al., 2022). One potential mechanism for this therapy may be the promotion of sense of purpose. Sense of purpose is characterized by engagement in personally meaningful activities that align with one's overarching life goals; it is plausible that by promoting movement in valued directions, behavioral activation may help increase sense of purpose. Therefore, while future research is needed, incorporating elements of behavioral activation into CMT support groups and other programs could promote sense of purpose and help people better cope with anxiety and discrimination associated with their disorder which could lead to improvements in well-being.

Second, policies and practices aimed at promoting sense of purpose could be implemented in clinical care. One practice that may be beneficial for people with CMT is regular assessment of sense of purpose. Although those with CMT regularly undergo testing of strength and physical functioning, evaluation of sense of purpose and overall psychological well-being is not a standard care practice. However, this study supports prior research suggesting that CMT may negatively impact well-being (Burns et al., 2010; Taniguchi et al., 2013; Vinci et al., 2005). While more research is needed to determine causality, the results of this study suggest that improvement of sense of purpose may be possible through intervention on psychosocial factors. Assessing sense of purpose and psychosocial risk factors in people with CMT at routine care appointments may be beneficial and allow for early intervention. In addition, while the results suggest that CMT may negatively impact sense of purpose, many participants reported feeling fairly purposeful on average. Although there are no cutoff values for high versus low sense of purpose, past work has suggested the importance of considering

above the midpoint as an important threshold for measures of purpose and meaning (Heintzelman & King, 2014). As such, it is noteworthy that this sample aligns with the norm from past work insofar that they were above the midpoint on average. Therefore, it is important for clinical care team members to avoid equating disability with distress, and potentially inadvertently conveying hopelessness or incapability to their patient, both of which may negatively impact sense of purpose. Instead, encouraging patients with CMT to pursue valued activities and helping them find solutions to engagement barriers may help promote sense of purpose. While this discussion has explored a few potential implications of this work for clinical care, future research is needed to examine how physicians and other care team members can best promote sense of purpose and support the overall psychological well-being of their patients with CMT.

Limitations

Although this study provides valuable initial findings for research with the CMT community, there are limitations that should be considered. First, because this was an observational study, causation and effect directionality could not be determined. While our results suggest that psychosocial factors such as self-efficacy could have a direct impact on well-being, it is impossible to determine if a causal relationship exists between these variables. Second, this study only assessed participants at one timepoint, making it difficult to examine day-to-day well-being or how well-being changes over time. Third, this study was conducted in the middle of a global pandemic; the unique challenges presented by this health crisis may have impacted participants' usual resources and well-being. Fourth, since the CMT-related anxiety, CMT/disability community perceived support, and adaptive device measures were created for this study, future research is needed to further examine validity and reliability. Fifth, while our study assessed physical functioning, which helps serve as a proxy for CMT severity, CMT severity was not directly assessed. Therefore, it is difficult to determine the impact this may have had on our results. Sixth, since participants reported on their own well-being and experiences, self-rating biases may have influenced the results. Future research incorporating observer reports is needed to examine whether these biases had any impact on the results.

Constraints on Generality

While we attempted to increase diversity in our sample by recruiting nationally and utilizing multiple recruitment platforms, limiting factors within our sample demographics may impact the generalizability of these results. First, the majority of the sample was middle-to-older adults; therefore, it is unclear whether these findings hold in younger adults or children with CMT. Second, this study was conducted with adults residing in the United States. Therefore, it is unknown whether these results translate to people with CMT living outside of the United States. Third, while CMT is found in all racial and ethnic groups (Bösenberg & Larkin, 2006), our sample was predominantly White. Therefore, future research is needed to determine if these findings extend to individuals with other racial or ethnic identities.

Conclusion

This study examined the relationships between psychosocial factors and sense of purpose in adults with CMT, as well as the role of sense of purpose as a moderator. Results suggest that many

psychosocial factors are strongly associated with sense of purpose in this population. In addition, sense of purpose may be beneficial for promoting well-being in individuals with CMT. These results can raise awareness in the CMT, medical, and scientific communities of the impact psychosocial factors may have on well-being, and to inform future care and interventions for those with CMT. As one of the first studies examining psychological well-being in people with CMT, this study lays the groundwork and provides valuable directions for future research on how to promote sense of purpose among this population.

References

- Alim, T. N., Feder, A., Graves, R. E., Wang, Y., Weaver, J., Westphal, M., Alonso, A., Aigbogun, N. U., Smith, B. W., Doucette, J. T., Mellman, T. A., Lawson, W. B., & Charney, D. S. (2008). Trauma, resilience, and recovery in a high-risk African-American population. *American Journal of Psychiatry*, *165*(12), 1566–1575. <https://doi.org/10.1176/appi.ajp.2008.07121939>
- Amtmann, D., Bamer, A. M., Cook, K. F., Askew, R. L., Noonan, V. K., & Brockway, J. A. (2012). University of Washington self-efficacy scale: A new self-efficacy scale for people with disabilities. *Archives of Physical Medicine and Rehabilitation*, *93*(10), 1757–1765. <https://doi.org/10.1016/j.apmr.2012.05.001>
- Börsbo, B., Gerdle, B., & Peolsson, M. (2010). Impact of the interaction between self-efficacy, symptoms and catastrophising on disability, quality of life and health in with chronic pain patients. *Disability and Rehabilitation*, *32*(17), 1387–1396. <https://doi.org/10.3109/09638280903419269>
- Bösenberg, A., & Larkin, K. (2006). Anaesthesia and Charcot–Marie–Tooth disease: Syndromic vignettes in anaesthesia. *Southern African Journal of Anaesthesia and Analgesia*, *12*(4), 131–133. <https://doi.org/10.1080/22201173.2006.10872453>
- Boswell, J. F., Iles, B. R., Gallagher, M. W., & Farchione, T. J. (2017). Behavioral activation strategies in cognitive-behavioral therapy for anxiety disorders. *Psychotherapy*, *54*(3), 231–236. <https://doi.org/10.1037/pst0000119>
- Bousquet, J., Knani, J., Dhivert, H., Richard, A., Chicoye, A., Ware, J. E., & Michel, F. B. (1994). Quality of life in asthma. I. Internal consistency and validity of the SF-36 questionnaire. *American Journal of Respiratory and Critical Care Medicine*, *149*(2), 371–375. <https://doi.org/10.1164/ajrccm.149.2.8306032>
- Boyle, P. A., Barnes, L. L., Buchman, A. S., & Bennett, D. A. (2009). Purpose in life is associated with mortality among community-dwelling older persons. *Psychosomatic Medicine*, *71*(5), 574–579. <https://doi.org/10.1097/PSY.0b013e3181a5a7c0>
- Britt-Spells, A. M., Slebodnik, M., Sands, L. P., & Rollock, D. (2018). Effects of perceived discrimination on depressive symptoms among black men residing in the United States: A meta-analysis. *American Journal of Men's Health*, *12*(1), 52–63. <https://doi.org/10.1177/1557988315624509>
- Bronk, K. C., Hill, P. L., Lapsley, D. K., Talib, T. L., & Finch, H. (2009). Purpose, hope, and life satisfaction in three age groups. *The Journal of Positive Psychology*, *4*(6), 500–510. <https://doi.org/10.1080/17439760903271439>
- Bronk, K. C., & Mitchell, C. (2022). Considering purpose through the lens of prospection. *The Journal of Positive Psychology*, *17*(2), 281–287. <https://doi.org/10.1080/17439760.2021.2016899>
- Burns, J., Ryan, M. M., & Ouvrier, R. A. (2010). Quality of life in children with Charcot–Marie–Tooth disease. *Journal of Child Neurology*, *25*(3), 343–347. <https://doi.org/10.1177/0883073809339877>
- Burrow, A. L., Hill, P. L., & Sumner, R. (2016). Leveling mountains: Purpose attenuates links between perceptions of effort and steepness. *Personality and Social Psychology Bulletin*, *42*(1), 94103. <https://doi.org/10.1177/0146167215615404>

- Charcot-Marie-Tooth Association. (2021). *What is Charcot-Marie-Tooth disease (CMT)?* Charcot-Marie-Tooth Association. <https://www.cmtausa.org/understanding-cmt/what-is-cmt/>
- Chung, H., Kim, J., Park, R., Bamer, A. M., Bocell, F. D., & Amtmann, D. (2016). Testing the measurement invariance of the University of Washington Self-Efficacy Scale short form across four diagnostic subgroups. *Quality of Life Research*, 25(10), 2559–2564. <https://doi.org/10.1007/s11136-016-1300-z>
- Cohen, R., Bavishi, C., & Rozanski, A. (2016). Purpose in life and its relationship to all-cause mortality and cardiovascular events: A meta-analysis. *Psychosomatic Medicine*, 78(2), 122–133. <https://doi.org/10.1097/PSY.0000000000000274>
- Cordeiro, J. L. C., Marques, W., Hallak, J. E. C., & Osório, F. L. (2014). Charcot-Marie-Tooth disease, psychiatric indicators and quality of life: A systematic review. *ASN Neuro*, 6(3), Article AN20130048. <https://doi.org/10.1042/AN20130048>
- Crego, A., Yela, J. R., Gómez-Martínez, M.Á., Riesco-Matías, P., & Petisco-Rodríguez, C. (2021). Relationships between mindfulness, purpose in life, happiness, anxiety, and depression: Testing a mediation model in a sample of women. *International Journal of Environmental Research and Public Health*, 18(3), Article 925. <https://doi.org/10.3390/ijerph18030925>
- deRoon-Cassini, T. A., de St. Aubin, E., Valvano, A., Hastings, J., & Horn, P. (2009). Psychological well-being after spinal cord injury: Perception of loss and meaning making. *Rehabilitation Psychology*, 54(3), 306–314. <https://doi.org/10.1037/a0016545>
- Diener, E., Emmons, R. A., Larsen, R. J., & Griffin, S. (1985). The Satisfaction with Life Scale. *Journal of Personality Assessment*, 49(1), 71–75. https://doi.org/10.1207/s15327752jpa4901_13
- Dimidjian, S., Barrera, M., Martell, C., Muñoz, R. F., & Lewinsohn, P. M. (2011). The origins and current status of behavioral activation treatments for depression. *Annual Review of Clinical Psychology*, 7(1), 1–38. <https://doi.org/10.1146/annurev-clinpsy-032210-104535>
- Hackett, R. A., Steptoe, A., Lang, R. P., & Jackson, S. E. (2020). Disability discrimination and well-being in the United Kingdom: A prospective cohort study. *BMJ Open*, 10(3), Article e035714. <https://doi.org/10.1136/bmjopen-2019-035714>
- Heintzelman, S. J., & King, L. A. (2014). Life is pretty meaningful. *American Psychologist*, 69(6), 561–574. <https://doi.org/10.1037/a0035049>
- Heisel, M. J., & Flett, G. L. (2004). Purpose in life, satisfaction with life, and suicide ideation in a clinical sample. *Journal of Psychopathology and Behavioral Assessment*, 26(2), 127–135. <https://doi.org/10.1023/B:JOBA.0000013660.22413.e0>
- Hill, P. L., & Burrow, A. L. (2012). Viewing purpose through an Eriksonian lens. *Identity*, 12(1), 74–91. <https://doi.org/10.1080/15283488.2012.632394>
- Hill, P. L., Burrow, A. L., & Bronk, K. C. (2016). Persevering with positivity and purpose: An examination of purpose commitment and positive affect as predictors of grit. *Journal of Happiness Studies*, 17(1), 257–269. <https://doi.org/10.1007/s10902-014-9593-5>
- Hill, P. L., Edmonds, G. W., & Hampson, S. E. (2019). A purposeful lifestyle is a healthful lifestyle: Linking sense of purpose to self-rated health through multiple health behaviors. *Journal of Health Psychology*, 24(10), 1392–1400. <https://doi.org/10.1177/1359105317708251>
- Hill, P. L., Sin, N. L., Turiano, N. A., Burrow, A. L., & Almeida, D. M. (2018). Sense of purpose moderates the associations between daily stressors and daily well-being. *Annals of Behavioral Medicine*, 52(8), 724–729. <https://doi.org/10.1093/abm/kax039>
- Hill, P. L., & Turiano, N. A. (2014). Purpose in life as a predictor of mortality across adulthood. *Psychological Science*, 25(7), 1482–1486. <https://doi.org/10.1177/0956797614531799>
- Hooker, S. A., & Masters, K. S. (2016). Purpose in life is associated with physical activity measured by accelerometer. *Journal of Health Psychology*, 21(6), 962–971. <https://doi.org/10.1177/1359105314542822>
- Hooker, S. A., Slattengren, A. H., Boyle, L., & Sherman, M. D. (2020). Values-based behavioral activation for chronic pain in primary care: A pilot study. *Journal of Clinical Psychology in Medical Settings*, 27(4), 633–642. <https://doi.org/10.1007/s10880-019-09655-x>
- Horowitz, B. P., & Vanner, E. (2010). Relationships among active engagement in life activities and quality of life for assisted-living residents. *Journal of Housing for the Elderly*, 24(2), 130–150. <https://doi.org/10.1080/02763891003757056>
- Hsu, H.-C., Chang, C.-J., Tung, H.-H., & Wang, T.-J. (2019). Disability, emotional distress and well-being among patients with lumbar spondylo-lysthesis. *Journal of Clinical Nursing*, 28(21–22), 3866–3878. <https://doi.org/10.1111/jocn.14992>
- Kim, E. S., Chen, Y., Nakamura, J. S., Ryff, C. D., & VanderWeele, T. J. (2022). Sense of purpose in life and subsequent physical, behavioral, and psychosocial health: An outcome-wide approach. *American Journal of Health Promotion*, 36(1), 137–147. <https://doi.org/10.1177/08901171211038545>
- Kim, E. S., Delaney, S. W., & Kubzansky, L. D. (2019). Sense of purpose in life and cardiovascular disease: Underlying mechanisms and future directions. *Current Cardiology Reports*, 21(11), Article 135. <https://doi.org/10.1007/s11886-019-1222-9>
- Kim, E. S., Kawachi, I., Chen, Y., & Kubzansky, L. D. (2017). Association between purpose in life and objective measures of physical function in older adults. *JAMA Psychiatry*, 74(10), 1039–1045. <https://doi.org/10.1001/jamapsychiatry.2017.2145>
- Kim, E. S., Strecher, V. J., & Ryff, C. D. (2014). Purpose in life and use of preventive health care services. *Proceedings of the National Academy of Sciences*, 111(46), 16331–16336. <https://doi.org/10.1073/pnas.1414826111>
- Kim, M., Yeom, H.-E., & Jung, M. S. (2022). Validation and psychometric properties of the multidimensional scale of perceived social support among Korean breast cancer survivors. *Asia-Pacific Journal of Oncology Nursing*, 9(4), 229–235. <https://doi.org/10.1016/j.apjon.2022.01.004>
- Krieger, N., Smith, K., Naishadham, D., Hartman, C., & Barbeau, E. M. (2005). Experiences of discrimination: Validity and reliability of a self-report measure for population health research on racism and health. *Social Science and Medicine*, 61(7), 1576–1596. <https://doi.org/10.1016/j.socscimed.2005.03.006>
- Lee, Y. H., Liu, Z., Fatori, D., Bauermeister, J. R., Luh, R. A., Clark, C. R., Bauermeister, S., Brunoni, A. R., & Smoller, J. W. (2022). Association of everyday discrimination with depressive symptoms and suicidal ideation during the COVID-19 pandemic in the All of Us Research Program. *JAMA Psychiatry*, 79(9), Article 898. <https://doi.org/10.1001/jamapsychiatry.2022.1973>
- Lins, L., & Carvalho, F. M. (2016). SF-36 total score as a single measure of health-related quality of life: Scoping review. *SAGE Open Medicine*, 4, Article 2050312116671725. <https://doi.org/10.1177/2050312116671725>
- Lyons, R. A., Perry, I. M., & Littlepage, B. N. C. (1994). Evidence for the validity of the Short-form 36 Questionnaire (SF-36) in an Elderly Population. *Age and Ageing*, 23(3), 182–184. <https://doi.org/10.1093/ageing/23.3.182>
- McKnight, P. E., & Kashdan, T. B. (2009). Purpose in life as a system that creates and sustains health and well-being: An integrative, testable theory. *Review of General Psychology*, 13(3), 242–251. <https://doi.org/10.1037/a0017152>
- Mekawi, Y., Carter, S., Packard, G., Wallace, S., Michopoulos, V., & Powers, A. (2022). When (passive) acceptance hurts: Race-based coping moderates the association between racial discrimination and mental health outcomes among Black Americans. *Psychological Trauma: Theory, Research, Practice, and Policy*, 14(1), 38–46. <https://doi.org/10.1037/tra0001077>
- Moorer, P., Suurmeijer, T., Th, P. B. M., Foets, M., & Molenaar, I. W. (2001). Psychometric properties of the RAND-36 among three chronic disease (multiple sclerosis, rheumatic diseases and COPD) in the Netherlands. *Quality of Life Research*, 10(7), 637–645. <https://doi.org/10.1023/A:1013131617125>
- Nearchou, F., Davies, A., & Hennessy, E. (2022). Psychometric evaluation of the multi-dimensional scale of perceived social support in young adults with chronic health conditions. *Irish Journal of Psychological Medicine*, 39(4), 386–390. <https://doi.org/10.1017/ipm.2019.54>

- Pavot, W., & Diener, E. (2009). Review of the satisfaction with life scale. In E. Diener (Ed.), *Assessing well-being: The collected works of Ed Diener* (pp. 101–117). Springer. https://doi.org/10.1007/978-90-481-2354-4_5
- Pfund, G. N., Ratner, K., Allemand, M., Burrow, A. L., & Hill, P. L. (2022). When the end feels near: Sense of purpose predicts well-being as a function of future time perspective. *Aging and Mental Health, 26*(6), 1178–1188. <https://doi.org/10.1080/13607863.2021.1891203>
- Pinar, R. (2005). Reliability and construct validity of the SF-36 in Turkish cancer patients. *Quality of Life Research, 14*(1), 259–264. <https://doi.org/10.1007/s11136-004-2393-3>
- Ryff, C. D. (1989). Happiness is everything, or is it? Explorations on the meaning of psychological well-being. *Journal of Personality and Social Psychology, 57*(6), 1069–1081. <https://doi.org/10.1037/0022-3514.57.6.1069>
- Ryff, C. D., & Keyes, C. L. M. (1995). The structure of psychological well-being revisited. *Journal of Personality and Social Psychology, 69*(4), 719–727. <https://doi.org/10.1037/0022-3514.69.4.719>
- Scheier, M. F., Wrosch, C., Baum, A., Cohen, S., Martire, L. M., Matthews, K. A., Schulz, R., & Zdzienicka, B. (2006). The life engagement test: Assessing purpose in life. *Journal of Behavioral Medicine, 29*(3), 291–298. <https://doi.org/10.1007/s10865-005-9044-1>
- Shy, M. E., & Rose, M. R. (2005). Charcot-Marie-Tooth disease impairs quality of life: Why? And how do we improve it? *Neurology, 65*(6), 790–791. <https://doi.org/10.1212/01.wnl.0000181027.21574.df>
- Silverman, A. M., Verrall, A. M., Alschuler, K. N., Smith, A. E., & Ehde, D. M. (2017). Bouncing back again, and again: A qualitative study of resilience in people with multiple sclerosis. *Disability and Rehabilitation, 39*(1), 14–22. <https://doi.org/10.3109/09638288.2016.1138556>
- Taniguchi, J. B., Elui, V. M. C., Osório, F. L., Hallak, J. E. C., Crippa, J. A. S., Machado-de-Sousa, J. P., Kebbe, L. M., Lourenço, C. M., Scarel-Caminaga, R. M., & Marques Jr, W. (2013). Quality of life in patients with Charcot-Marie-Tooth disease type 1A. *Arquivos de Neuro-Psiquiatria, 71*(6), 392–396. <https://doi.org/10.1590/0004-282X20130045>
- Thompson, N. J., Coker, J., Krause, J. S., & Henry, E. (2003). Purpose in life as a mediator of adjustment after spinal cord injury. *Rehabilitation Psychology, 48*(2), 100–108. <https://doi.org/10.1037/0090-5550.48.2.100>
- Ulmer, A., Range, L. M., & Smith, P. C. (1991). Purpose in life: A moderator of recovery from bereavement. *OMEGA—Journal of Death and Dying, 23*(4), 279–289. <https://doi.org/10.2190/33VU-BANW-C54V-W7VF>
- Verduin, P. J. M., de Bock, G. H., Vliet Vlieland, T. P. M., Peeters, A. J., Verhoef, J., & Otten, W. (2008). Purpose in life in patients with rheumatoid arthritis. *Clinical Rheumatology, 27*(7), 899–908. <https://doi.org/10.1007/s10067-007-0822-8>
- Vinci, P., Serrao, M., Millul, A., Deidda, A., De Santis, F., Capici, S., Martini, D., Pierelli, F., & Santilli, V. (2005). Quality of life in patients with Charcot-Marie-Tooth disease. *Neurology, 65*(6), 922–924. <https://doi.org/10.1212/01.wnl.0000176062.44360.49>
- Ware, J. E., Gandek, B., & Group, I. P. (1994). The SF-36 health survey: Development and use in mental health research and the IQOLA Project. *International Journal of Mental Health, 23*(2), 49–73. <https://doi.org/10.1080/00207411.1994.11449283>
- Ware, J. E., & Sherbourne, C. D. (1992). The MOS 36-item short-form health survey (SF-36): I. Conceptual framework and item selection. *Medical Care, 30*(6), 473–483. <https://doi.org/10.1097/00005650-199206000-00002>
- Weston, S. J., Hill, P., & Mroczek, D. (2019). *Purpose in life is associated with health outcomes among diabetic adults*. <https://doi.org/10.31234/osf.io/4snbt>
- Williams, D. R., & Mohammed, S. A. (2009). Discrimination and racial disparities in health: Evidence and needed research. *Journal of Behavioral Medicine, 32*(1), 20–47. <https://doi.org/10.1007/s10865-008-9185-0>
- Williams, D. R., Yan, Y., Jackson, J. S., & Anderson, N. B. (1997). Racial differences in physical and mental health: Socio-economic status, stress and discrimination. *Journal of Health Psychology, 2*(3), 335–351. <https://doi.org/10.1177/135910539700200305>
- Zimet, G. D., Dahlem, N. W., Zimet, S. G., & Farley, G. K. (1988). The multidimensional scale of perceived social support. *Journal of Personality Assessment, 52*(1), 30–41. https://doi.org/10.1207/s15327752jpa5201_2

(Appendices follow)

Appendix A

CMT-Related Anxiety Scale

Scale:

- 1 = *none*
- 2 = *a small amount*
- 3 = *a moderate amount*
- 4 = *quite a bit*
- 5 = *a lot*

1. How much anxiety do you experience about tripping/falling due to your CMT?
2. How much anxiety do you experience about not being able to keep up with others due to your CMT?
3. How much anxiety do you experience about the physical inaccessibility of places you may visit?
4. How much anxiety do you experience about receiving judgment or negative reactions from others due to your CMT?
5. How much anxiety do you experience about potential equipment malfunctions (wheelchairs, braces, etc.)?
6. How much anxiety do you experience about the progressive nature of your CMT?

Avoidance Due to CMT-Related Anxiety

How often do you avoid situations or activities due to anxiety related to your CMT symptoms (fear of tripping, not being able to keep up, fear of judgment, etc.)?

- 1 = *never*
- 2 = *rarely*
- 3 = *sometimes*
- 4 = *often*
- 5 = *always*

Ranked Anxiety

Please rank the following by how much anxiety you experience (1 = *the thing you experience the most anxiety about*; 6 = *the thing you experience the least anxiety about*).

- _The progressive nature of your CMT
- _Tripping/falling
- _Being unable to keep up with others
- _Judgment or negative reactions from others regarding your CMT
- _Equipment malfunctions (wheelchair, braces, etc.)
- _The physical inaccessibility of places you may visit

Appendix B

CMT/Disability Community Perceived Support Scale

Scale:

- 1 = *strongly disagree*
- 2 = *disagree*
- 3 = *neutral*
- 4 = *agree*
- 5 = *strongly agree*

Please indicate the extent of your agreement with the following statements:

1. I feel a sense of connection to others with CMT.
2. I feel a sense of belonging within the CMT community.
3. I feel a sense of belonging within the greater disability community.
4. I have someone in my life who is willing and able to assist me with physical tasks when/if I need it.
5. I have someone in my life who I can talk to about CMT-related challenges or experiences.

Received December 10, 2022
 Revision received May 11, 2023
 Accepted July 29, 2023 ■