

# An exploratory study of exercise behaviours and barriers to participation in people with Charcot-Marie-Tooth disease: a focus on resistance training

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The aim of this study was to explore and describe the exercise behaviours and barriers in people with Charcot-Marie-Tooth disease (CMT), with a particular focus on resistance training (RT). Ninety-four Australian adults with a diagnosis of CMT completed an online survey. Fifty-seven percent of respondents reported performing some form of RT each week. Those performing RT engaged in more aerobic activity ( $P \leq 0.01$ ) and were involved in longer periods of structured exercise ( $P < 0.01$ ) compared to those not performing RT. The RT group was more likely to perceive their exercise levels as acceptable ( $P < 0.01$ ), that following a program was important ( $P = 0.02$ ), and that exercise is beneficial ( $P = 0.04$ ). The RT group were more likely to have been advised to

exercise ( $P = 0.02$ ). Common barriers to exercise were fatigue (64.9%), pain (57.4%), motivation (51.1%), and time (46.8%). RT status did not influence the type of barriers experienced. Weekly RT time was positively associated with exercise satisfaction ( $r = 0.43$ ,  $P < 0.01$ ) and walking distance prior to resting ( $r = 0.29$ ,  $P = 0.04$ ). The findings suggest that positive exercise experiences, advice, assistance from a trainer, and potentially greater resources may influence participation in RT for people with CMT.

**Keywords:** Muscle strength, Neurodegeneration, Peripheral neuropathy, Physical activity, Exercise adherence

## INTRODUCTION

Charcot-Marie-Tooth disease (CMT) is one of the most common motor and sensory neurological disorders. It affects an estimated 1 in 2,500 individuals (Szigeti and Lupski, 2009), equating to approximately 10,144 cases in Australia. These figures are conservative given the likelihood of under diagnosis. CMT is inherited through a mutation of genes responsible for the structure and function of peripheral nerves. Both motor and sensory nerves are affected, predominantly in the lower limbs. The most common type of CMT is 1A, accounting for roughly 60% of total CMT cases (Fridman et al., 2015). Onset of CMT generally occurs during childhood or within the first 20 years of life. Damage to the peripheral nerves causes muscle wasting, weakness, and sensory loss in the extremities, with a distal to proximal progression (Maggi et al., 2011; Newman et al., 2007). Depending on severity, the disease can have a signif-

icant impact on functional ability, particularly due to muscle weakness in the foot and ankle. Activities such as walking and jogging become increasingly difficult and presents a higher risk of ankle sprains and falls (Burns et al., 2009). These limitations lead to a loss of independence and reduced health-related quality of life (Pfeiffer et al., 2001). Currently, no curative treatments are available for CMT and because it is a highly variable disease, identifying specific management procedures is challenging (Pisciotta et al., 2021).

Exercise has been identified as an effective treatment strategy for people with CMT (Corrado et al., 2016). A systematic review by Sman et al. (2015) investigated the effects of exercise in this clinical population and found that most studies involved resistance training (RT) interventions. The results from this review showed that RT was effective for improving strength, functional activities, and muscle fibre size. Similarly, the studies that involved aerobic training showed favourable changes in some measures of strength

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and functional activities, as well as an increase in aerobic capacity (El Mhandi et al., 2008). There were also studies that used combined exercise interventions with positive changes found for ankle flexibility, balance, and physical function (Maggi et al., 2011; Matjacić and Zupan, 2006). The greater proportion of studies examining RT compared to other exercise modalities in people with CMT is most likely due its ability to target specific muscle weaknesses (Voet et al., 2013). However, most exercise studies involving this clinical population generally involve small sample sizes, which presents a problem when analysing the data and interpreting the results. Additionally, there are difficulties with making inferences to the broader CMT population, hence preventing the exercise findings from being generalized.

For people with CMT, exercise intolerance and undue fatigue are common complaints (Mori et al., 2020). This reduced physical ability is directly related to the disease, but physical deconditioning is also a contributing factor. As a result, exercise might not be enjoyable for some people with CMT, which could negatively affect long-term adherence to exercise programs. There has been one study to date that has explored the perceived facilitators and barriers to physical activity in people with CMT (Anens et al., 2015). This study surveyed 44 people with CMT and found that facilitators of physical activity were self-efficacy for physical activity, activity-related factors, and assistive devices. Additionally, self-efficacy for physical activity and fatigue explained 32% of the variation in physical activity. Physical activity refers to any movement that a person performs, while exercise is defined as planned, structured, repetitive, and intentional movement that is intended to improve or maintain physical fitness (Caspersen et al., 1985). Currently no study to date has investigated the exercise habits and barriers to exercise in people with CMT.

The aim of this study was to explore and describe the exercise behaviours and barriers in people with CMT. Since RT is an important exercise modality for people with CMT, a particular focus was placed on the results of respondents involved in RT compared to respondents that did not participate in this exercise modality. The findings may be helpful to allied health practitioners when prescribing exercise for people with CMT.

## MATERIALS AND METHODS

### Participants

Individuals aged 20 to 76 years with a diagnosis of CMT and residing in Australia participated in this study. One hundred forty-one people commenced the survey, however there were 94 in-

dividuals that completed it (i.e., 66.7% completion rate). An online survey was created using Research Electronic Data Capture (REDCap) digital and was hosted on The University of Sydney's REDCap server. The survey was advertised through social media networks and on the CMT Australia website. Additionally, people with CMT from previous studies at The University of Sydney were invited via SMS to complete the survey. Interested individuals were provided with details of a landing page to check study eligibility and for access to the survey link. The eligibility criteria included being clinically diagnosed with CMT, aged  $\geq 18$  years, and living in Australia. The survey was available from June to October 2022. Each subject read and signed (using a checkbox) an informed consent document approved by the University of Sydney Human Research Ethics Committee (approval number: 2022/277).

### Design

The survey consisted of 30 questions and was divided into four subgroups. These subgroups included participant information, exercise and medical history, current exercise habits, and other habits and potential barriers. Specific questions included in the survey are shown in Table 1. The survey questions were mostly fixed response (with some open-ended). Additionally, for questions that asked about satisfaction with physical activity levels in the past year and quantifying weekly exercise volume a slider scale was used (e.g., 0 to 100). Respondents gauged their intensity of an aerobic activity based on descriptors. Low intensity allowed engaging in conversation to be easy, moderate-intensity activity made engaging in conversation harder than normal, and high-intensity activity made engaging in conversation difficult. Prior to releasing the survey, it was pilot tested to assess for errors and evaluation of user experience. All surveys were screened for duplicates and questionable responses (e.g., untruthful responses indicated by unrealistic respondent characteristics).

### Data analysis

Statistical analyses were performed using IBM SPSS Statistics ver. 28.0 (IBM Co., Armonk, NY, USA). Normality of data was assessed using the Kolmogorov–Smirnov test. Given the inconsistent normal data distribution, nonparametric tests were used for all analyses. Continuous data was presented as median with interquartile range. Differences between RT and non-RT (NRT) groups for continuous data were analysed using the Mann–Whitney *U*-test. Categorical data was presented as number of responses and percentage of respondents. The differences between RT and NRT groups for categorical data was assessed using the chi-square test

of independence. Relationships between RT time (per week) and other variables with continuous data were assessed with the Spearman rank correlation coefficient. Both the strength of relation-

**Table 1.** Survey questions

1. Participant information
1.1 What is your age?
1.2 What is your height?
1.3 What is your weight?
1.4 Have you been diagnosed with any other medical conditions?
1.5 If you are taking any medications, please select all that apply.
1.6 How many years since your diagnosis of CMT?
1.7 What health care professionals have been involved in your care for CMT?
2. Exercise and medical history
2.1 Rate the severity of your CMT diagnosis.
2.2 What symptoms potentially related to CMT do you experience regularly?
2.3 Have you participated in a structured exercise program within the past year?
2.4 Have you been advised in the past to participate in an exercise program?
2.5 How satisfied are you with your physical activity levels during the past year?
2.6 Have your exercise habits changed during the COVID-19 pandemic?
3. Current exercise habits
3.1 In a typical week, how many minutes do you spend participating in low-intensity aerobic activity?
3.2 In a typical week, how many minutes do you spend participating in moderate-intensity aerobic activity?
3.3 In a typical week, how many minutes do you spend participating in high-intensity aerobic activity?
3.4 In a typical week, how many minutes do you spend participating in resistance training?
3.5 Who do you usually exercise with?
3.6 What locations do you exercise at?
3.7 If you are currently following a program, who supplied this to you?
3.8 Do you believe your current aerobic activity levels are acceptable for the management of CMT?
3.9 Do you believe your current resistance training activity levels are acceptable for the management of CMT?
3.10 If you have participated in exercise during the last year, have you noticed improvements in your overall health and wellbeing?
3.11 At any stage have you been concerned that participating in exercise may have a negative impact on your health and wellbeing?
3.12 If you have participated in a structured exercise program during the past year, is there anything you would have changed?
4. Other habits and potential barriers
4.1 How physically strenuous is your job?
4.2 How psychologically strenuous is your job?
4.3 How long can you walk before needing to rest?
4.4 What are some barriers that might prevent you from participating in a structured exercise program?
4.5 How important do you think it is for you to be participating in a structured exercise program?

COVID-19, coronavirus disease 2019; CMT, Charcot-Marie-Tooth disease.

ships and 95% confidence interval (CI) were reported. Strength of correlations were qualitatively assessed using the following criteria: trivial ( $r < 0.1$ ), small ( $r > 0.1$  to  $0.3$ ), moderate ( $r > 0.3$  to  $0.4$ ), strong ( $r > 0.5$  to  $0.7$ ), very strong ( $r > 0.7$  to  $0.9$ ), nearly perfect ( $r > 0.9$ ), and perfect ( $r = 1.0$ ) (Hopkins et al., 2009). Significance was set at  $P < 0.05$ .

## RESULTS

### Background characteristics

The characteristics of the respondents are presented in Table 2. Most respondents reported having at least one comorbidity (53.2%) and taking one or more medications (74.4%). The most common comorbidities reported were related to musculoskeletal conditions (RT,  $n = 9$ , NRT,  $n = 9$ ), mental health (RT,  $n = 10$ , NRT,  $n = 6$ ), neurological disorders (RT,  $n = 3$ , NRT,  $n = 7$ ), and respiratory disease (RT,  $n = 6$ , NRT,  $n = 4$ ). Other comorbidities reported were related to cardiovascular disease (RT,  $n = 2$ , NRT,  $n = 6$ ), metabolic conditions (RT,  $n = 3$ , NRT,  $n = 5$ ), digestive conditions (RT,  $n = 2$ , NRT,  $n = 6$ ), cancer (RT,  $n = 3$ , NRT,  $n = 1$ ), and renal disease (RT,  $n = 1$ , NRT,  $n = 1$ ). Respondents in RT compared to NRT reported taking a greater number of medications ( $P = 0.04$ ). Respondents had a median time since CMT diagnosis of 20.5 years and rated the severity of their CMT symptoms as moderate (median of 61.5 on 100-point scale). The most common CMT symptoms reported were weakness (93.6%), decreased sensation in feet (85.1%), fatigue (83.0%), trips and falls (78.7%), pain (77.7%), and tingling sensation (61.7%).

Most respondents were employed (64.9%), with 38.3% of respondents involved in sedentary type occupations. Of the respondents employed, 23.0% perceived the physical nature of their job as being moderately to very strenuous, while 68.9% perceived the psychological nature of their job as being moderately to very strenuous. Difficulties with activities of daily living (ADL) were report-

**Table 2.** Description of background variables for total sample and differences between groups that did and did not engage in resistance training

Variable	All (n=94)	RT (n=54)	NRT (n=40)	P-value
Age (yr)	52.0 (45.0–62.0)	50.5 (45.8–62.3)	53.5 (45.0–61.8)	0.81
BMI (kg/m <sup>2</sup> )	27.2 (23.9–32.8)	26.3 (23.9–32.3)	29.1 (23.7–33.7)	0.22
Total comorbidities (n)				
1–2	77	45	32	0.11
3–4	14	9	5	
≥5	3	0	3	

(Continued)

**Table 2.** Description of background variables for total sample and differences between groups that did and did not engage in resistance training (Continued)

Variable	All (n=94)	RT (n=54)	NRT (n=40)	P-value
Total medications (n)				
1–2	57	34	23	0.81
3–4	25	13	12	
≥5	12	7	5	
Years since diagnosis	20.5 (10.0–34.3)	20.0 (10.0–33.3)	27.0 (12.0–39.0)	0.32
CMT severity (0–100)	61.0 (50.0–75.3)	62.5 (50.0–75.0)	60.0 (50.0–76.0)	0.64
CMT symptoms (n)				
Pain				
Yes	73	42	31	0.98
No	21	12	9	
Weakness				
Yes	88	49	39	0.19
No	6	5	1	
Fatigue				
Yes	78	43	35	0.32
No	16	11	5	
Tingling sensations				
Yes	58	31	27	0.32
No	36	23	13	
Trips or falls				
Yes	74	42	32	0.80
No	20	12	8	
Difficulty concentrating				
Yes	26	17	9	0.34
No	68	37	31	
Difficulty with ADLs				
Yes	48	30	18	0.31
No	46	24	22	
Poor sleep				
Yes	48	28	20	0.86
No	46	26	20	
Decreased sensation in legs/feet				
Yes	80	43	37	0.08
No	14	11	3	
Difficulties with ADLs (n)				
Yes	83	49	34	0.39
No	11	5	6	
ADLs difficult frequency (n)				
Never	5	2	3	0.18
Occasionally	14	11	3	
Frequently	75	41	34	
Distance walking before rest (min)	20 (10–52.5)	25.0 (13.8–60.0)	15.0 (6.3–30.0)	0.06

Values are presented as median (interquartile range) or number. RT, resistance training; NRT, no resistance training; BMI, body mass index; CMT, Charcot-Marie-Tooth disease; ADL, activities of daily living.

ed by 88.3% of respondents with these difficulties being experienced frequently (79.8% of respondents). Respondents reported being able to walk for a median time of 20 min prior to needing a rest. Most respondents had physiotherapists involved in their care (78.7%), with no difference been RT and NRT groups. Exercise physiologists were part of the care team for 42.6% of respondents, with a greater number of respondents in the RT group compared to NRT group using this allied health service (51.9% versus 30.0%, respectively,  $P = 0.03$ ). There were no other statistical differences between the RT and NRT groups for background characteristics.

### Exercise habits and related perceptions

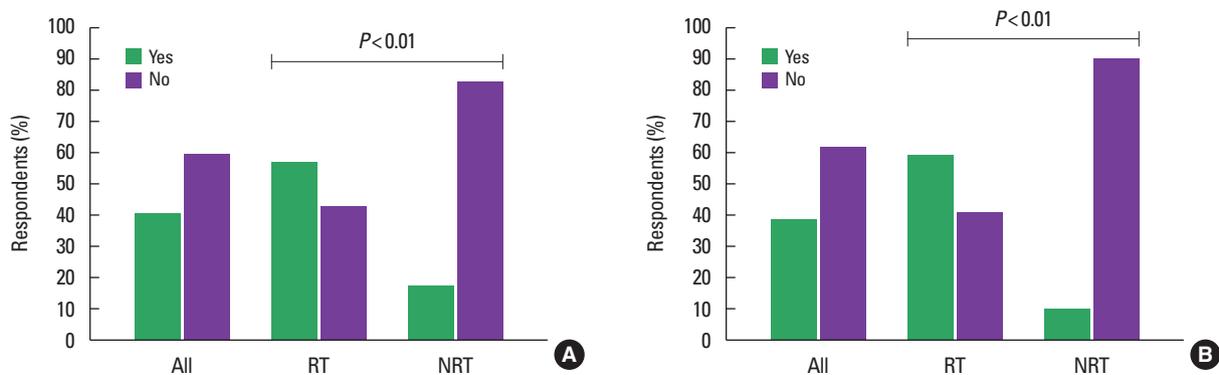
Respondents in the RT group reported engaging in a median time of 57 min of RT weekly. The RT group compared to NRT group also engaged in a greater amount of aerobic training at low, moderate, and high intensities in a typical week ( $P \leq 0.01$ ) (Table 3). Just less than half of all respondents (48.9%) had completed structured exercise training for a period of greater than 3 months within the previous year. There was a significant difference between the exercise history of the RT and NRT groups ( $P < 0.01$ ). No structured exercise training was performed by 65% of respondents in the NRT group, while 72.2% of the respondents in the RT group reported performing greater than 3 months of structured exercise training. Most respondents (61.7%) had been advised in the past to exercise to improve their health, with a greater number in the RT compared to NRT group (72.2% vs. 47.5%, respectively,  $P = 0.02$ ). Approximately 40% of respondents perceived their level of resistance and aerobic training was acceptable, with more respondents expressing this perception in the RT group ( $P < 0.01$ ) (Fig. 1A, B). Belief in the benefits of exercise was reported by 67% of respondents, with this view higher amongst the RT group. A slightly lower than moderate exercise satisfaction was reported, with a higher satisfaction among the RT group compared to the NRT group ( $P < 0.01$ ). Most respondents considered following a structured exercise program very important (68.0%), with more respondents in the RT group having a similar opinion ( $P = 0.02$ ). Of the respondents that exercised, 79.7% noticed an improvement in their health and wellbeing, with a significantly greater number among the RT group ( $P = 0.03$ ).

Most respondents exercised with someone (69.1%), with a trainer being the most popular support (54.1%), followed by exercising with friends or family (16.0%), and exercising in a group (13.8%). Respondents in the RT compared NRT group were more likely to use a trainer (48.1% vs. 17.0%;  $P = 0.02$ ), with no other differenc-

**Table 3.** Exercise habits and related perceptions for total sample and differences between groups that did and did not engage in resistance training

Variable	All (n=94)	RT (n=54)	NRT (n=40)	P-value
Resistance training (min)	19.5 (0-66.0)	57.0 (30.0-132.0)	0	<0.01
Aerobic training (min)				
Low intensity	90.0 (30.0-180.8)	120.0 (60.0-186.8)	57.0 (21.0-150.0)	0.01
Moderate intensity	21.0 (0-75.8)	34.5 (11.3-120.0)	0 (0-29.3)	<0.01
High intensity	0 (0-16.5)	4.5 (0-44.3)	0	<0.01
Advised to exercise				
Yes	58	39	19	0.02
No	36	15	21	
Belief of benefit from exercise				
Yes	63	44	19	0.04
No	22	10	12	
Belief of negative impact from exercise				
Yes	34	18	16	0.24
No	55	36	19	
Exercise history satisfaction	43.0 (25.0-67.0)	54.5 (30.0-83.5)	29.0 (15.3-50.0)	<0.01
Importance of following an exercise program				
Not important	2	1	1	0.02
Somewhat important	28	10	18	
Very important	64	43	21	
Noticed improvements in health and wellbeing from exercise				
Yes	47	38	9	0.03
No	12	6	6	

Values are presented as median (interquartile range) or number. RT, resistance training; NRT, no resistance training.



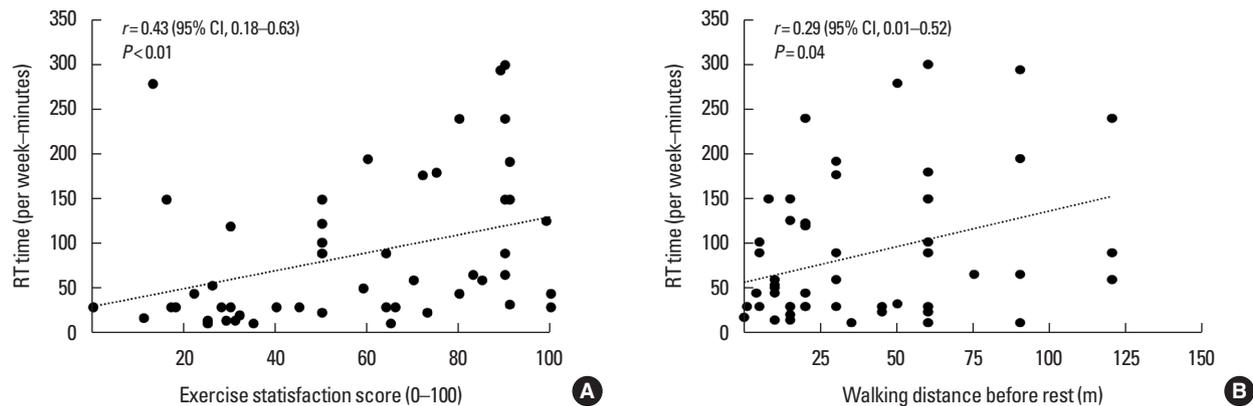
**Fig. 1.** Perception of acceptable level of aerobic training and resistance training. (A) Responses for aerobic training. (B) Responses for resistance training. All, total sample; RT, resistance training; NRT, no resistance training.

es between groups for exercise support. Exercising at home was performed by 58.5% of respondents, followed by in a park (48.9%), and at an exercise clinic or facility (28.7%). Respondents in the RT compared to the NRT group were more likely to exercise at a clinic or facility (44.4% vs. 7.5%, respectively;  $P < 0.01$ ), with no significant differences between groups for the other exercise locations. For the RT group, 46.2% of respondents received their ex-

ercise program from a health professional or trainer.

### Effect of COVID-19 on exercise habits and barriers to exercise

As a result of the coronavirus disease 2019 (COVID-19) pandemic, exercise habits changed for 59.6% of respondents, with reduced motivation being the main factor (28.7%). Other factors affecting



**Fig. 2.** Relationships between resistance training time (per week), exercise satisfaction, and estimated walking distance before rest. (A) Resistance training (RT) time and exercise satisfaction. (B) RT time and walking distance before rest.

exercise habits included limited access to equipment and facilities (22.3%), and changes in health (17.0%). Limited access to equipment and facilities had a greater impact on RT compared to NRT (31.5% vs. 10.0%,  $P = 0.01$ ). The most popular barriers to exercise included fatigue (64.9%), pain (57.4%), motivation (51.1%), time (46.8%), access to facilities (28.7%), access to equipment (27.7%), and transport (11.7%). There were no significant differences between the RT and NRT groups for barriers to exercise.

### Relationship between RT time (per week) and predictor variables

A moderate positive relationship was observed between weekly RT time and exercise history satisfaction ( $r = 0.43$ ,  $P < 0.01$ ; 95% CI, 0.18–0.63) (Fig. 2A). There was a small positive relationship found between RT time and walking distance prior to resting ( $r = 0.29$ ,  $P = 0.04$ ; 95% CI, 0.01–0.52) (Fig. 2B). No significant relationships were found between RT time and age ( $r = -0.26$ ,  $P = 0.06$ ; 95% CI, -0.50 to 0.02), years since diagnosis ( $r = -0.23$ ,  $P = 0.09$ ; 95% CI, -0.48 to 0.05), and CMT severity ( $r = -0.14$ ,  $P = 0.14$ ; 95% CI, -0.40 to 0.14).

## DISCUSSION

This study aimed to explore and describe the exercise behaviours and barriers in people with CMT, with a particular focus on respondents that did versus did not perform RT (i.e., RT versus NRT, respectively). Just more than half of the respondents reported performing some form of RT each week. The RT compared to NRT group engaged in more aerobic training and were involved in a longer period of structured exercise. There was a consistent theme in reported perceptions between groups. The RT group

was more likely to perceive their exercise levels as acceptable, that following a program was important, and that exercise is beneficial. It is also worth noting that respondents in the RT group compared to NRT group were more likely to have been advised to exercise. The most commonly reported barriers to exercise included fatigue, pain, motivation, time, access to facilities, access to equipment, and transport. However, RT status did not influence the type of barrier experienced. Weekly RT time was positively associated with exercise satisfaction and walking distance prior to resting. Findings from this study suggest that people with CMT that engage in RT have more positive expectations about exercise, which may assist them with overcoming any potential barriers they encounter. However, since an online survey was used to gather information for this study, caution is warranted when interpreting the findings as this method of data collection is considered to be of low quality.

The American College of Sports Medicine (ACSM) recommends that for health benefits adults achieve 500–1,000 MET min of aerobic exercise per week through 150 min at a moderate intensity, 75 min at a vigorous-intensity, or a combination of moderate and vigorous aerobic exercise (Garber et al., 2011). The respondents for the present study mostly performed low-intensity exercise of a relatively small volume (median of 90 min per week), therefore not meeting these guidelines. However, it should be noted that there are currently no specific exercise guidelines for adults with CMT. There was a large difference in the amount of aerobic exercise performed by the RT group compared to NRT group at all intensities. Aerobic training in people with CMT has been shown to improve physical function (El Mhandi et al., 2008) and cardiorespiratory fitness (Wallace et al., 2019). Additionally, people with CMT are less active compared to the general popula-

tion (Ramdharry et al., 2017), therefore usually present with aerobic deconditioning and secondary disuse muscle atrophy. The RT group reported they were able to walk for a median of 25 min before resting compared to a median of 15 min for the NRT group. However, a difference between groups just failed to reach statistical significance for this outcome ( $P = 0.06$ ). It is possible that respondents belonging to the RT group had better physical conditioning which allowed them to engage in more aerobic exercise.

The RT group engaged in approximately 1 hr of this RT per week. Unfortunately, the number of sessions per week was not captured in the survey, preventing a comparison to the ACSM RT recommendations. These guidelines state that healthy adults should engage in at least 2 days per week of RT (Garber et al., 2011). However, a recent meta-analysis found that the maximum risk reduction (approximately 10%–20%) for all-cause mortality, cardiovascular disease and total cancer was observed with approximately 30–60 min per week of RT (Momma et al., 2022). There were 10 respondents that reported performing 12–24 min of RT per week, while the remaining respondents from the RT group reported performing 30–300 min per week. This suggests that most of the RT group may be receiving health-related benefits from engaging this exercise modality. However, the amount of weekly RT does not indicate its effectiveness for people with CMT. Further information about the acute programming variables for RT such as intensity, volume, frequency, and exercise type would be required to determine whether the exercise being performed is optimal for improving muscle performance. For people with CMT, training specific muscle groups may provide greater benefit for function, gait, and balance. These might include resistance exercises targeting the abductor and adductor hip muscles (Hackett et al., 2021; Roberts-Clarke et al., 2016), dorsiflexor, and plantarflexor muscles (Burns et al., 2017). As for the respondents from the RT group who reported believing their exercise levels were insufficient, it is possible that their exercise prescription was suboptimal and therefore not achieving the desired results.

Moinuddin (2021) concluded that studies on RT in people with CMT have generally failed to find statistically significant benefits in terms of strength gain. This author speculated that RT can be difficult to implement and study in CMT due to the nature of the disease. For the respondents in the present study, the RT group reported more favourable attitudes towards engaging in exercise. The RT group adopted more frequent and longer lasting exercise, as well as being more likely to believe in benefits from exercising. These characteristics in the RT group are consistent with Bandura's theory concerning greater adherence to an activity, which is

thought to be due to higher expectations of both self-efficacy and outcome expectancy (Perkins et al., 2008). Additionally, exercise satisfaction was positively related to weekly RT time in agreement with the effects of positive psychological traits influencing involvement in RT. The only other significant predictor of RT time was walking distance prior to resting, suggesting that fitness or physical function plays a role.

Since a greater percentage of respondents in the RT group compared to NRT group were advised to participate in exercise by a health care professional, it appears that education was another factor that influenced exercise behaviours. Improving knowledge about how certain lifestyle changes can positively affect overall health is crucial for increasing self-efficacy and reducing helplessness (Collado-Mateo et al., 2021). Additionally, the person providing the advice or the exercise program may affect adherence. Higher levels of exercise adherence may be observed when prescribed by physicians compared to other professionals (e.g., personal trainers) (Collado-Mateo et al., 2021). This is consistent with the health belief model, where expected benefits relates to involvement in an activity (Khodaveisi et al., 2021). While 79.6% of the RT group reported believing that following an exercise program was very important, only 46% of this group received an exercise program from a health professional or trainer. Therefore, it appears that multiple factors are likely to be involved in the decision of respondents to engage in regular RT.

Most respondents were overweight with at least 1–2 comorbidities, were employed in sedentary type occupations, and had difficulties with ADLs on a regular basis. Although the reported severity of CMT was moderate, the overall health of respondents was generally poor. Findings from the systematic review conducted by Sman et al. (2015) reported mean body mass index  $\geq 29$  kg/m<sup>2</sup> for people with CMT that participated in exercise interventions. As results showed the most common CMT symptoms were weakness, decreased sensation in feet, fatigue, trips and falls, pain, and tingling sensations, the poor physical health of the respondents is likely attributed to increased sedentary behaviour influenced by these disabling symptoms.

While the RT group was satisfied with their level of exercise and more likely to notice improvements in their health and well-being during exercise, the outlook for the NRT group was less positive. The NRT group were more likely to have negative attitudes towards exercise, believed their current exercise levels were unacceptable, and were less satisfied with their physical activity levels. There were minimal statistical differences between the RT and NRT groups for the background characteristics and more im-

portantly, the barriers to exercise were similar for both groups. Therefore, it appears that the RT group may be more driven (i.e., motivated) to exercise, have more resources to assist with implementing exercise, and have greater support to continue exercising. However, the survey was inadequately designed to explain exercise barriers and this topic would be better explored using a qualitative study design (i.e., interviews and focus group discussions).

Most respondents participating in structured exercise reported being supervised by a trainer. No research to date has examined the effects of supervised versus unsupervised exercise training in people with CMT. However, in older adults, exercising under supervision is an effective strategy for improving adherence (Picorelli et al., 2014), as well as training-induced adaptations (Lacroix et al., 2016). Therefore, it would seem highly plausible that similar effects from supervised exercise would be achieved in people with CMT. It might not always be feasible for people with CMT to access to a trainer (due to location or finances). Survey results showed that the majority of respondents reported engaging in some form of exercise at home. There is limited evidence to support the feasibility and efficacy of home-based exercise programs, however, this is a strategy that may successfully address the barriers of time (46.8%) and transport (11.7%). There were also 44.4% of respondents in the RT group that reported being more likely to exercise at a clinic or facility. Provided adequate equipment is available, home-based programs may further improve RT participation. Furthermore, such a program may overcome changes to exercise habits caused by events such as the COVID-19 pandemic where these habits were negatively affected due to limited access to equipment and facilities.

There are certain limitations concerning this study that should be acknowledged. The web-based survey enabled a greater coverage of respondents, so that data could be generalizable to people with CMT. However, the findings from this study may not be generalizable to this clinical population. There was a 66.7% completion rate which meant that there was a 33.3% noncompletion bias. A total of 94 respondents completed the survey which was more than double the previous physical activity survey in people with CMT (Anens et al., 2015). Although, the sample size of this population would need to be larger to allow the results to be generalized to people with CMT. Other limitations with using the web-based survey tool included the lack of details or depth on exercise prescription, exercise barriers, and perceptions to exercise. Therefore, the addition of an interview (as mentioned previously) might have been a better method for collecting additional information as well as to clarify certain questions. Unfortunately, gen-

der was not captured in the survey which prevented further analysis of this factor on exercise behaviour and barriers to participation. Another issue relating to surveys in research is the accuracy of respondents' answers. The accuracy and truth of the responses for the present study is unknown. However, respondents were informed (i.e., statements placed on survey page) that all information was confidential and only available to the authors, which may have increased the accuracy and truth of the responses.

This study provides information on the exercise behaviours and barriers to participation of Australian adults with CMT. Approximately half of the respondents engaged in regular RT with these same respondents also having more positive attitudes towards exercise. Barriers to exercise participation were similar between respondents regardless of RT participation. Positive exercise experiences, advice, assistance from a trainer, and potentially greater resources may have influenced RT participation. However, further research using a qualitative study design is needed to examine this topic more thoroughly. Allied health professionals should consider the information covered in this study when prescribing exercise for people with CMT to assist with long-term adherence to a program.

## CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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## REFERENCES

- Anens E, Emtner M, Hellström K. Exploratory study of physical activity in persons with Charcot-Marie-Tooth disease. *Arch Phys Med Rehabil* 2015;96:260-268.
- Burns J, Ryan, MM, Ouvrier RA. Evolution of foot and ankle manifesta-

- tions in children with CMT1A. *Muscle Nerve* 2009;39:158-166.
- Burns J, Sman AD, Cornett KMD, Wojciechowski E, Walker T, Menezes MP, Mandarakas MR, Rose KJ, Bray P, Sampaio H, Farrar M, Refshauge KM, Raymond J. Safety and efficacy of progressive resistance exercise for Charcot-Marie-Tooth disease in children: a randomised, double-blind, sham-controlled trial. *Lancet Child Adolesc Health* 2017;1:106-113.
- Caspersen CJ, Powell KE, Christenson GM. Physical activity, exercise, and physical fitness: definitions and distinctions for health-related research. *Public Health Rep* 1985;100:126-131.
- Collado-Mateo D, Lavín-Pérez AM, Peñacoba C, Del Coso J, Leyton-Román M, Luque-Casado A, Gasque P, Fernández-Del-Olmo M, Amado-Alonso D. Key factors associated with adherence to physical exercise in patients with chronic diseases and older adults: an umbrella review. *Int J Environ Res Public Health* 2021;18:2023.
- Corrado B, Ciardi G, Bargigli C. Rehabilitation management of the Charcot-Marie-Tooth syndrome: a systematic review of the literature. *Medicine (Baltimore)* 2016;95:e3278.
- El Mhandi L, Millet GY, Calmels P, Richard A, Oullion R, Gautheron V, Féasson L. Benefits of interval-training on fatigue and functional capacities in Charcot-Marie-Tooth disease. *Muscle Nerve* 2008;37:601-610.
- Fridman V, Bundy B, Reilly MM, Pareyson D, Bacon C, Burns J, Day J, Feely S, Finkel RS, Grider T, Kirk CA, Herrmann DN, Laurá M, Li J, Lloyd T, Sumner CJ, Muntoni F, Piscoquito G, Ramchandren S, Shy R, Siskind CE, Yum SW, Moroni I, Pagliano E, Zuchner S, Scherer SS, Shy ME. CMT subtypes and disease burden in patients enrolled in the inherited neuropathies consortium natural history study: a cross-sectional analysis. *J Neurol Neurosurg Psychiatry* 2015;86:873-878.
- Garber CE, Blissmer B, Deschenes MR, Franklin BA, Lamonte MJ, Lee IM, Nieman DC, Swain DP. American College of Sports Medicine position stand. Quantity and quality of exercise for developing and maintaining cardiorespiratory, musculoskeletal, and neuromotor fitness in apparently healthy adults: guidance for prescribing exercise. *Med Sci Sports Exerc* 2011;43:1334-1359.
- Hackett D, Roberts-Clarke D, Halaki M, Burns J, Fiatarone Singh M, Fornusek C. High intensity power training in middle-aged women with Charcot-Marie-Tooth disease: a case series. *Int J Ther Rehabil* 2021; 28:1-12.
- Hopkins WG, Marshall SW, Batterham AM, Hanin J. Progressive statistics for studies in sports medicine and exercise science. *Med Sci Sports Exerc* 2009;41:3-13.
- Khodaveisi M, Azizpour B, Jadidi A, Mohammadi Y. Education based on the health belief model to improve the level of physical activity. *Phys Act Nutr* 2021;25:17-23.
- Lacroix A, Kressig RW, Muehlbauer T, Gschwind YJ, Pfenninger B, Bruegger O, Granacher U. Effects of a supervised versus an unsupervised combined balance and strength training program on balance and muscle power in healthy older adults: a randomized controlled trial. *Gerontology* 2016;62:275-288.
- Maggi G, Monti Bragadin M, Padua L, Fiorina E, Bellone E, Grandis M, Reni L, Bennicelli A, Grosso M, Saporiti R, Scorsoni D, Zuccharino R, Crimi E, Schenone A. Outcome measures and rehabilitation treatment in patients affected by Charcot-Marie-Tooth neuropathy: a pilot study. *Am J Phys Med Rehabil* 2011;90:628-637.
- Matjacić Z, Zupan A. Effects of dynamic balance training during standing and stepping in patients with hereditary sensory motor neuropathy. *Disabil Rehabil* 2006;28:1455-1459.
- Moinuddin A. Charcot-Marie-Tooth disease: genetic predisposition and effect of resistance training, endurance training, physical activity and orthosis in attenuating its severity. *Eur J Mol Clin Med* 2021;7:1333-1342.
- Momma H, Kawakami R, Honda T, Sawada SS. Muscle-strengthening activities are associated with lower risk and mortality in major non-communicable diseases: a systematic review and meta-analysis of cohort studies. *Br J Sports Med* 2022;56:755-763.
- Mori L, Signori A, Prada V, Pareyson D, Piscoquito G, Padua L, Pazzaglia C, Fabrizi GM, Picelli A, Schenone A. Treadmill training in patients affected by Charcot-Marie-Tooth neuropathy: results of a multicenter, prospective, randomized, single-blind, controlled study. *Eur J Neurol* 2020;27:280-287.
- Newman CJ, Walsh M, O'Sullivan R, Jenkinson A, Bennett D, Lynch B, O'Brien T. The characteristics of gait in Charcot-Marie-Tooth disease types I and II. *Gait Posture* 2007;26:120-127.
- Perkins JM, Multhaup KS, Perkins HW, Barton C. Self-efficacy and participation in physical and social activity among older adults in Spain and the United States. *Gerontologist* 2008;48:51-58.
- Pfeiffer G, Wicklein EM, Ratusinski T, Schmitt L, Kunze K. Disability and quality of life in Charcot-Marie-Tooth disease type 1. *J Neurol Neurosurg Psychiatry* 2001;70:548-550.
- Picorelli AM, Pereira LS, Pereira DS, Felício D, Sherrington C. Adherence to exercise programs for older people is influenced by program characteristics and personal factors: a systematic review. *J Physiother* 2014; 60:151-156.
- Pisciotta C, Saveri P, Pareyson D. Challenges in treating Charcot-Marie-Tooth disease and related neuropathies: current management and future perspectives. *Brain Sci* 2021;11:1447.
- Ramdharry GM, Pollard AJ, Grant R, Dewar EL, Laurá M, Moore SA, Hallsworth K, Ploetz T, Trenell MI, Reilly MM. A study of physical activity comparing people with Charcot-Marie-Tooth disease to nor-

- mal control subjects. *Disabil Rehabil* 2017;39:1753-1758.
- Roberts-Clarke D, Fornusek C, Saigal N, Halaki M, Burns J, Nicholson G, Fiatarone Singh M, Hackett D. Relationship between physical performance and quality of life in Charcot-Marie-Tooth disease: a pilot study. *J Peripher Nerv Syst* 2016;21:357-364.
- Sman AD, Hackett D, Fiatarone Singh M, Fornusek C, Menezes MP, Burns J. Systematic review of exercise for Charcot-Marie-Tooth disease. *J Peripher Nerv Syst* 2015;20:347-362.
- Szigeti K, Lupski JR. Charcot-Marie-Tooth disease. *Eur J Hum Genet* 2009; 17:703-710.
- Voet NB, van der Kooij EL, Riphagen II, Lindeman E, van Engelen BG, Geurts AC. Strength training and aerobic exercise training for muscle disease. *Cochrane Database Syst Rev* 2013;7:Cd003907.
- Wallace A, Pietrusz A, Dewar E, Dudzic M, Jones K, Hennis P, Sterr A, Baio G, Machado PM, Laurá M, Skorupinska I, Skorupinska M, Butcher K, Trenell M, Reilly M, Hanna M, Ramdharry G. Community exercise is feasible for neuromuscular diseases and can improve aerobic capacity. *Neurology* 2019;92:e1773-e1785.