Research Topics

Exploring physical activity and sedentary behaviour in physical disability

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Published in
Frontiers in Rehabilitation Sciences

ISSN 1664-8714
ISBN 978-2-83250-087-3
DOI 10.3389/978-2-83250-087-3

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Exploring physical activity and sedentary behaviour in physical disability

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Citation
Ryan, J., Kerr, C., Kilbride, C. B., Norris, M., eds. (2022). *Exploring physical activity and sedentary behaviour in physical disability*. Lausanne: Frontiers Media SA.
doi: 10.3389/978-2-83250-087-3

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frontiers | Frontiers in Rehabilitation Sciences

TYPE Editorial
PUBLISHED 17 August 2022
DOI 10.3389/fresc.2022.1006039

OPEN ACCESS

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SPECIALTY SECTION
This article was submitted to Disability, Rehabilitation, and Inclusion, a section of the journal Frontiers in Rehabilitation Sciences

RECEIVED 28 July 2022
ACCEPTED 02 August 2022
PUBLISHED 17 August 2022

CITATION
Ryan J, Kerr C, Kilbride C and Norris M (2022) Editorial: Exploring physical activity and sedentary behaviour in physical disability. Front. Rehabilit. Sci. 3:1006039. doi: 10.3389/fresc.2022.1006039

Editorial: Exploring physical activity and sedentary behaviour in physical disability

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Editorial on the Research Topic
Exploring physical activity and sedentary behaviour in physical disability

By Ryan J, Kerr C, Kilbride C, Norris M. (2022) Front. Rehabilit. Sci. 3:1006039. doi: 10.3389/fresc.2022.1006039

Increasing physical activity and reducing sedentary behaviour reduces the risk of premature mortality, cardiovascular disease, cancer, depression, and type 2 diabetes. For children and adults with physical disabilities (1–5), benefits on function and community participation may also accrue (6, 7). On average, children and adults with physical disabilities are less active than people without disabilities and have higher levels of sedentary behaviour (5, 8). Guidelines recommend that adults with disabilities participate in at least 150 min of moderate activity per week and children participate in at least 60 min of moderate activity daily (6, 7). Supporting children and adults with physical disabilities to increase physical activity and reduce sedentary behaviour may enhance community participation, improve health, and reduce health and social inequalities (6, 7). However, influences on physical activity and sedentary behaviour are multi-faceted and interdependent (9, 10). This research topic aimed to explore the interactions between individual, social and structural factors that influence physical activity participation and sedentary behaviour among children and adults with physical disabilities. In doing so, we aimed to further knowledge and understanding of associations between physical activity, sedentary behaviour, community participation, and physical, mental, and social wellbeing among people with physical disabilities (including the impact of societal and physical barriers). We also aimed to showcase innovative policy and practice approaches to enhancing physical activity and reducing sedentary behaviours across the lifespan.

The nine papers included in the research topic were authored by teams from the United Kingdom, United States of America, Australia and the Netherlands. They employed a variety of research designs, and considered various facets of physical activity in adults with mobility disability Morgan et al. (2022), stroke Church et al. (2021), multiple sclerosis Stennett et al. (2021); Lavelle et al. (2022); Fortune et al. (2021) and rare neurological conditions Ramdharry et al. (2021); and in children and young people with physical disabilities Bolster et al. (2021); Sharma et al. (2021); Sansare et al. (2021). Collectively, this research topic provides a snapshot of the breadth and diversity of research in the area and highlights some of the key considerations when developing, implementing and evaluating interventions to increase physical activity for people with disabilities of all ages.

The International Classification of Functioning, Disability and Health (ICF) (11) was employed explicitly or implicitly in many of the papers in this research topic as a framework for describing the experiences and impact of living with physical disability, operationalising domains that interventions might target, and considering outcomes of physical activity interventions. Three papers highlighted the complexity, interdependency and potentially fluctuating nature of the physical, psychological, participatory and contextual challenges faced by this population when considering, or participating in, physical activity Stennett et al. (2021); Ramdharry et al. (2021); Bolster et al. (2021). Detailed consideration of contextual factors when developing physical activity interventions, potentially before function and disability factors, was highlighted in a number of papers Morgan et al. (2022); Stennett et al. (2021); Ramdharry et al. (2021); Bolster et al. (2021); Sharma et al. (2021). Interestingly, Ramdharry et al noted a mismatch between outcome tools reported in the literature, which focused primarily on activity, compared to the participation-focused outcomes of importance articulated by people with rare neurological conditions Ramdharry et al. (2021). Church et al. noted a similar trend in their paper, demonstrating that although body structure and function outcomes were measured in all 15 empirical studies in their rapid review of high intensity training in people with stroke, participation outcomes were only measured in four Church et al. (2021). Taken together, the papers in this research topic use the language of the ICF to articulate the many influences on physical activity. They advocate for theory-driven physical activity interventions that incorporate behaviour change components, take due cognisance of the individual's health status, their environment and their individual goals, and evaluate outcomes of importance to the individual.

Measurement was a strong theme in the papers included in this research topic. As detailed above, an ICF approach was often employed with a strong focus on participation outcomes. However, measurement validity was also addressed. Lavelle et al. demonstrated poor criterion validity of commercially available devices to monitor step-count and activity time in people with multiple sclerosis Lavelle et al. (2022). In addition to the issues this may cause when evaluating effectiveness of physical activity interventions, it also resulted in frustration and distrust amongst wearers, which could potentially negatively impact motivation to be physically active. It appears that there is still a need for development of psychometrically robust, user-friendly methods of objective measurement of physical activity in people with disabilities.

Sustaining participation in physical activity can be challenging and may be strongly influenced by both personal and environmental factors. Sharma et al. used technology to overcome environmental barriers, demonstrating the feasibility and acceptability of an online physical activity intervention for people aged 12–21 years with a physical disability Sharma et al. (2021). In contrast, Morgan et al. reported outcomes from a long-running community-based exercise programme delivered in an accessible community facility Morgan et al. (2022). Bolster et al. also strongly advocated for consideration of the environment in which physical activity interventions are delivered but acknowledged that provision of physical activity "therapy" in the everyday environment is logistically difficult and thus costly within current service delivery models Bolster et al. (2021). This suggests that innovation is required at policy and health systems levels to deliver impactful interventions in a cost-effective manner.

The value of increasing physical activity and reducing sedentary behaviour for everyone is undisputed – we know "why" it is important. For people with disabilities, the "who", "what", "where", "when" and "how" to optimise physical activity participation are still up for discussion. This research topic demonstrates this complexity but also the innovation and variety in design, methods, implementation and evaluation of physical activity interventions for people with disabilities. It also provides a stimulus for further research in this important area.

Author contributions

JR led the conception of the research topic with contributions from MN, CK and CK. All authors contributed to analysis and interpretation. CK drafted the manuscript. All authors contributed to the article and approved the submitted version.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

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ORIGINAL RESEARCH
published: 30 April 2021
doi: 10.3389/fresc.2021.651688

An Online Physical Activity Intervention for Youth With Physical Disabilities: A Pilot Study

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Background: Physical activity (PA) interventions are limited in number and reach for youth with physical disabilities (YPD) who experience systemic barriers that may preclude their in-person participation. Further, a lack of theory in the development and evaluation of PA interventions impedes our understanding and replication of active components of behavior change. These limitations pose challenges in the effective promotion of PA in YPD. Theory-based and more inclusive methods of PA intervention delivery must be explored in our efforts to promote PA and overall health in YPD.

Methods: A pilot study was conducted to evaluate the feasibility and outcomes of an online, 4-week social cognitive theory-based PA intervention for YPD. Intervention feasibility (implementation fidelity, intervention compliance, and intervention acceptability) was evaluated through manual documentation, weekly feedback questionnaires, and open-ended feedback at 1-month post-intervention. Targeted social cognitive (outcome expectations, self-efficacy [task, self-regulatory, barrier] and self-regulation) and PA behavior outcomes were self-reported at baseline and 1-week and 1-month post-intervention.

Results: Sixteen YPD ($M_{age} = 17.4 \pm 2.7$ years, 69% female) completed the study. Intervention feasibility was supported by high implementation fidelity (100%), high intervention compliance (>90%), and positive ratings on indicators of acceptability for all weeks of the intervention (weekly feedback questionnaire means ranging from 5.74 to 6.19 out of 7). Through open-ended feedback, participants indicated the intervention was easy to use and understand, favorably shifted their self-awareness and personal meaning of PA, and provided value and potential for future use pertaining to the learned self-regulation skills and strategies. Participants also provided formatting and content recommendations for intervention improvement. Repeated measures ANOVAs showed significant and large effect sizes for changes in participants' task ($p = 0.01$, $n^2p = 0.28$) and barrier ($p = 0.02$, $n^2p = 0.24$) self-efficacy, goal-setting and planning and scheduling behaviors ($ps < 0.001$, $n^2ps = 0.42$), and self-reported PA behavior ($p = 0.02$, $n^2p = 0.26$).

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Specialty section:
This article was submitted to Disability, Rehabilitation, and Inclusion, a section of the journal Frontiers in Rehabilitation Sciences

Received: *10 January 2021*
Accepted: *31 March 2021*
Published: *30 April 2021*

Citation:
Sharma R, Latimer-Cheung AE, Cairney J and Arbour-Nicitopoulos KP (2021) An Online Physical Activity Intervention for Youth With Physical Disabilities: A Pilot Study. Front. Rehabilit. Sci. 2:651688. doi: 10.3389/fresc.2021.651688

Conclusions: An online PA intervention for YPD is feasible and may offer potential benefit through the enhancement of self-efficacy, self-regulation, and PA behavior. Continued research is necessary to understand the efficacy and longer-term outcomes of online, theory-based interventions for YPD as a PA promotion strategy.

Keywords: youth, physical disabilities, physical activity, online intervention, intervention—behavioral, social cognitive theory

INTRODUCTION

Nearly 4% of Canadian children and youth under the age of 15 years and 13% of Canadian youth and young adults ages 15–24 years have a disability causing daily activity limitations (1, 2). Specifically, youth with physical disabilities (YPD) are at increased risk of experiencing health disparities and developing secondary health conditions related to the presence of health risk behaviors, such as insufficient physical activity (PA) (3). Despite the physical and psychosocial benefits that YPD can experience through participating in PA (4), YPD are reported to be less physically active than their typically developing peers (5) largely due to the presence of barriers to participation (6). Exploring strategies to promote PA during adolescence and young adulthood is critical, as the early adoption of positive health behaviors facilitates the maintenance of those behaviors into and throughout adulthood (7).

Existing PA interventions for YPD are limited in number and in their use of theory (8). This poses challenges for the effective promotion of PA and reinforces the need for theory-based interventions for YPD. Utilizing theory in the development and evaluation of behavior change interventions is critical for identifying and replicating the active components of an intervention leading to potential behavior change, with one of the most prominent theories in PA promotion research being social cognitive theory (SCT). Key SCT constructs that aid in our understanding and promotion of PA behavior include outcome expectations, self-efficacy, and self-regulation (e.g., goal-setting, planning, and self-monitoring) (9). According to SCT, self-efficacy is a direct determinant of behavior and has indirect effects on behavior through its influence on expectations of positive outcomes and the increased use of self-regulation strategies that are essential to achieve and maintain behavior change (9). Although limited, there is evidence supporting the use of SCT in PA interventions for youth with visual impairments (10) and cerebral palsy (11), such that youth demonstrated an increase in at least one of the targeted SCT constructs (i.e., outcome expectations, self-efficacy, and self-regulation) and in their PA behavior, respectively. Despite the absence of maintenance effects in both studies, this early evidence of short-term social cognitive and behavioral change in youth with visual impairments and cerebral palsy demonstrates that SCT may be an appropriate theoretical framework to inform the development of a PA intervention for YPD and warrants its continued exploration.

Despite the importance of theory in the development of behavior change interventions, the presence of environmental barriers to participation (e.g., inaccessible facilities, lack of available transportation) (6) also necessitates consideration of the mode of intervention delivery. The Internet may serve as an appropriate delivery alternative by eliminating environmental barriers that may preclude the participation of YPD in face-to-face PA interventions. Although online interventions have well-documented challenges related to attrition (12), their accessibility, increasingly sophisticated capabilities, and potential to engage YPD warrants further attention. Given the novelty of online theory-based PA interventions for YPD, a pilot study is necessary to understand whether future large-scale implementation could be practical and elicit meaningful change (13). Thus, this pilot study aimed to explore: (1) the feasibility of an online SCT-based PA intervention for YPD targeting outcome expectations, self-efficacy, and self-regulation; and (2) short- and longer-term changes in the targeted social cognitive and PA behavior outcomes as an initial assessment of potential intervention impact (13). Given the pilot nature of this study, hypotheses were not tested (13).

MATERIALS AND METHODS

Intervention Design and Development

A 4-week intervention ("*Plan to Move*") was developed by the first author and delivered on a web-hosting service called Weebly©. The structure and content of *Plan to Move* was guided in part by an existing, evidence- and SCT-based PA program for inactive adults with spinal cord injury (14). This existing program was chosen as a guide given its focus on similar SCT constructs (e.g., self-efficacy, self-regulation) and demonstrated efficacy in increasing leisure-time PA in a segment of the population with a physical disability (14). Thus, each week of the current intervention targeted a separate SCT construct, such that Weeks 1 through 4 targeted outcome expectations, task self-efficacy, self-regulation, and barrier self-efficacy, respectively.

Each week of *Plan to Move* consisted of an online session and corresponding independent activity. The content of the independent activities was adapted from the abovementioned guiding intervention and included age-appropriate modifications to the language and examples used. During these independent activities, for example, participants were asked to identify personally relevant benefits of engaging in PA (outcome expectations), reflect on positive PA experiences (task

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Abbreviations: PA, physical activity; LTPAQ-SCI, Leisure-Time Physical Activity Questionnaire for People with Spinal Cord Injury; RM-ANOVA, repeated measures analyses of variance; SCT, social cognitive theory; YPD, youth with physical disabilities.

self-efficacy), engage in goal-setting, planning, and self-monitoring (self-regulation), and establish a coping plan (barrier self-efficacy).

Participants received access to and completed from home each week's online session and corresponding independent activity sequentially. Each online session included multiple webpages that participants clicked to progress through, with YouTube™ videos embedded to supplement in-text information. These embedded videos were in the format of whiteboard voiceover animations and were specifically developed for this intervention by the first author using an online video animation software called Raw Shorts. The videos varied in length, ranging from 1:00 to 2:50, in minutes and seconds (m:ss), and provided salient examples of the topics introduced in the online sessions (e.g., goal-setting and planning for PA). A manipulation check, in the form of a knowledge-based question, was included on the final webpage of each week's session. To reinforce content and encourage participants to apply the learned skills and strategies, the independent activities were to be completed after the online sessions. Each week of the intervention (i.e., online session and independent activity) was designed to be completed within ∼20–25 min.

Prior to enrolling participants, adjustments (e.g., audio of YouTube™ videos, activity formatting) were made to the intervention based on feedback from two YPD in a pre-testing phase. Weekly intervention content, including the content of the YouTube™ videos, is detailed in **Appendix A**.

Participants

Participants were recruited from an existing database of YPD who participated in past PA research and from a provincial organization that provides programs and services to YPD using electronic recruitment flyers. Inclusion criteria were: (a) self-reported having a physical disability; (b) aged 12–21 years, with the upper threshold of the age range reflecting the transition age (18–21 years) for youth with disabilities from child to adult rehabilitation and education services in Ontario, Canada (15); (c) able to read and speak in English; and (d) able to complete surveys over the telephone. Due to the nature of participation, exclusion criteria were: (a) self-reported visual, hearing, and/or cognitive impairment; and/or (b) participation in elite-level sport. As per the National Institutes of Health's (NIH) guidelines for feasibility and pilot studies (13), a power analysis was not conducted. Institutional research ethics approval was obtained prior to recruitment (Protocol #33624).

Study Procedure

After confirming eligibility, informed parental consent and youth assent were provided via telephone for participants under the age of 18. Youth over the age of 18 provided informed consent. Consent and assent were documented by the researcher. Next, participants completed a demographics questionnaire and baseline assessment of the targeted social cognitive and PA behavior outcomes via telephone with the researcher delivering the intervention. The next day, participants received the link to the first online session and the corresponding independent activity in Word document format. After completing the first online session, indicated by the completion of the embedded manipulation check, participants were required to complete and return the independent activity to the researcher to gain access to the link and independent activity for Week 2. The same access-restricted procedure was followed for Weeks 3 and 4. The researcher sent text message reminders to participants if there was no indication of engagement with the online session or independent activity 5 days after receiving intervention materials or once they reached the threshold for "late" completion of the respective week of the intervention. Other than the researcher's involvement in the delivery of intervention materials, delivery of text message reminders, and availability to troubleshoot technical issues or answer questions, participants' engagement in the intervention was entirely self-led. Participants were scheduled to complete the same assessment that was administered at baseline with the same researcher via telephone 1 week and 1 month after completing the Week 4 online session and independent activity. The administration of the baseline and post-intervention assessments took ∼30–45 min to complete.

Measures

Demographics

Participants reported their age, gender, ethnicity, height, weight, disability type and duration, and use of a mobility device.

Intervention Feasibility

Aligning with the NIH Framework for Developing and Testing Mind and Body Interventions (13), feasibility was conceptualized as: (a) *implementation fidelity (dose and adherence)*, (b) *intervention compliance*, and (c) *intervention acceptability*. *Implementation fidelity* (i.e., delivered *dose*) and *intervention compliance* (i.e., online session and independent activity completion) were dichotomized as "complete" or "incomplete" for each week. "Complete" indicated that the session was delivered by the researcher to the participant (*dose*) and the manipulation check and independent activity were completed by the participant (*intervention compliance*). *Adherence* to the delivery schedule (i.e., each week was delivered within 7 days of participants completing the previous week) and participants' timely completion of the online sessions and independent activities (i.e., within 7 days of receiving access) were dichotomized as "on-time" or "late". Mean view time duration in minutes and seconds (m:ss) and mean view time percentage of each YouTube™ video was extracted from YouTube™ Analytics. *Intervention acceptability* was evaluated through feedback questionnaires completed by participants at the end of each week's independent activity. Participants rated on a Likert scale of 1 (strongly disagree) to 7 (strongly agree) whether each week of the intervention was: interesting, easy to understand, taught them new and trustworthy information, easy to navigate, and presented information and strategies that were helpful (16). Weekly acceptability scores were calculated as means of participants' ratings of these parameters. For additional detail on intervention acceptability, participants provided open-ended feedback during the 1-month post-intervention assessment on program satisfaction, utility, potential impact, and recommended improvements.

Social Cognitive and Behavioral Outcomes

Outcome Expectations

Participants completed an 11-item adjectival instrument on their personal beliefs about particular outcomes occurring as a result of engaging in PA and the value they place on those particular outcomes (e.g., "Physical activity will help me to have an adventure," and "Being adventurous is fun.") (17). Items were rated on a scale of 1 (never) to 6 (always). This scale has demonstrated acceptable reliability among typically developing youth (17). To reduce participant burden, the measure was truncated from 23 to 11 item pairs to only include outcome expectations that were targeted in the intervention.

Task Self-Efficacy

Participants completed a 7-item instrument on their context-specific confidence to engage in PA (e.g., "I can be physically active during my free time on most days."). Items were rated on a scale of 1 (disagree a lot) to 5 (agree a lot). This instrument has demonstrated acceptable test-retest reliability and factorial validity in typically developing youth (18). While this measure has been used to assess task self-efficacy for PA in youth with CP (11), psychometric properties were not specified.

Self-Regulatory Efficacy

Two components of self-regulatory efficacy were evaluated: goal-setting self-efficacy and planning and scheduling self-efficacy. To examine goal-setting self-efficacy, participants completed a 4-item instrument on their confidence in their ability to set PA goals in the next 4 weeks (e.g., "How confident are you that you can set realistic goals for maintaining your physical activity for the next 4 weeks?") (21). To examine planning and scheduling self-efficacy, participants completed a 7-item instrument on their confidence in their ability to schedule a self-managed PA routine in the next 4 weeks (e.g., "How confident are you that you can arrange your schedule to do physical activity each week no matter what for the next 4 weeks?") (21). Items from both instruments were rated from 0 (not at all confident) to 100 (completely confident). The language of both instruments was modified, such that "independent physical activity" was replaced with "physical activity." Both instruments have demonstrated acceptable reliability and validity in adults with spinal cord injury (20).

Barrier Self-Efficacy

Participants completed an 8-item scale on their confidence to overcome barriers that may prevent them from engaging in PA (e.g., "Assuming you are very motivated, how confident are you that you could participate in physical activity if you feel tired?"). Items were rated on a scale of 1 (not confident at all) to 7 (completely confident). Six of these eight items have demonstrated high internal consistency in typically developing children and youth (19). Two additional items relating to transportation problems and a lack of support were included, as these are salient PA barriers that individuals with physical disabilities often encounter (6). These items have demonstrated acceptable internal consistency in adults with spinal cord injury (20).

Self-Regulation Behavior

Two self-regulation behaviors were evaluated: goal-setting, and planning and scheduling. Participants completed the 10-item Exercise Goal Setting Scale (EGS) (22) and the 10-item Exercise Planning and Scheduling Scale (EPS) (22). Items from both the EGS and EPS were rated on a scale of 1 (does not describe) to 5 (describes completely). Examples from the EGS and EPS include, respectively: "I have developed a series of steps for achieving my physical activity goals," and "Physical activity is generally not a high priority when I plan my schedule." The EGS and EPS have demonstrated good internal reliability among college-aged youth (22). The language of the EGS and EPS was modified by replacing "exercise" with "physical activity."

Physical Activity

Participants completed the 6-item Leisure-Time Physical Activity Questionnaire for People with Spinal Cord Injury (LTPAQ-SCI), where they were asked to self-report the number of days and minutes on those days spent engaging in mild-, moderate-, and heavy-intensity PA, during their leisure time in the past 7 days (23). Weekly minutes of mild-, moderate-, and heavy-intensity PA were summed for a total amount of weekly minutes of PA overall. The LTPAQ-SCI has been found to be valid and reliable for persons with physical disabilities (23, 24).

Statistical Analysis

Statistical analyses for quantitative data were performed using SPSS Version 24.0. Descriptive statistics were conducted to summarize participants' demographic characteristics and quantitative feasibility outcomes measured through manual documentation and weekly feedback questionnaires. One-way repeated measures analyses of variance (RM-ANOVAs) were performed to assess social cognitive and PA behavior change from baseline to 1-week and 1-month post-intervention. Model assumptions were tested and the Greenhouse-Geisser correction was used for violations of the within-subjects assumption of homoscedasticity. Bonferroni corrections were performed to determine significant change(s) between the three time points. Given the absence of a power analysis, effect sizes were also used in the interpretation of results, such that η^2ps of 0.01, 0.06, and 0.14 represented small, medium, and large effect sizes, respectively (25).

Open-ended feedback from the 1-month post-intervention assessment was transcribed verbatim, de-identified through the assignment of pseudonyms and removal of identifying information, and underwent content analysis (26) by the first author. Transcripts were coded inductively to establish categories within the four topics, which were agreed upon by a critical friend (KPAN). Disagreements in the labeling of emerging categories or coding were resolved through discussion leading to consensus. The frequency of key words and phrases pertaining to each category was recorded.

TABLE 1 | Participant characteristics (*N* = 16).

Characteristic	Value
Age (years)	
M (SD)	17.4 (2.7)
Range	13–21
Sex, *n*	
Male	5
Female	11
Body Mass Index (kg/m^2), M (SD)	21.92 (6.55)
Ethnicity, *n*	
White	11
East Asian	2
Other (Black, South Asian, West Asian)	3
Type of Physical Disability, *n*	
Cerebral palsy	3
Muscular dystrophy	3
Neuromuscular disorder	2
Spinal cord injury	4
Other (brain injury, stroke, genetic disorder)	4
Years Living with Physical Disability, M (SD)	11.2 (6.7)
Use a Mobility Device, *n*	14
Manual wheelchair	4
Power wheelchair	5
Cane	2
Crutches	1
Other	2

M, mean; SD, standard deviation.

RESULTS

Participant Flow and Characteristics

Of the 33 eligible youth, two declined to participate (6%), 11 (33%) did not respond to the follow-up email, and 20 (61%) enrolled in the study. Sixteen youth (80%) completed the study in its entirety and four (20%) were lost to follow-up (data excluded from analyses). These four participants did not respond to emails regarding their continued participation in the study. **Table 1** provides participants' demographic characteristics.

Intervention Feasibility

Table 2 summarizes implementation fidelity and intervention compliance outcomes.

Implementation Fidelity

The intervention was delivered in its full *dose* (100%), meaning all online sessions and independent activities were delivered to all participants. Concerning *adherence*, all 4 weeks were delivered within an average of 4.35 (SD = 0.41) days after completion of the previous week's online session and independent activity. There was one instance of late delivery due to technical difficulties. In addition, the baseline and 1-week and 1-month post-intervention assessments were administered to all participants (100%). Adherence to the delivery schedule of the 1-week and 1-month post-intervention assessments was achieved, but was constrained by scheduling challenges in 3 of 16 participants. Overall, the intervention and assessments were delivered as intended.

Intervention Compliance

Overall, 13 of 16 participants completed all of the online sessions, indicated by the completion of the embedded manipulation check. In addition, 15 of 16 participants completed all of the independent activities. Out of the 64 total instances of intervention delivery to all participants (4 weeks x 16 participants), the overall percentage of completion of the online sessions and independent activities was 94% and 98%, respectively. Participants took, on average, 5.28 (SD = 4.02) days to complete each week of the intervention upon receiving access. Overall, 81% of the total delivered weeks of the intervention were completed on-time. Instances of "late" completion occurred between Weeks 2 and 4, with four participants not completing the respective online session and/or independent activity within 7 days of receiving access. On instances of "late" completion, participants took, on average, 4.33 (SD = 2.58) additional days to complete the online session and/or independent activity.

Participants viewed, on average, 69% of the total minutes (7:33 of 11:00) of the YouTube™ videos. The average view time percentages of each of the seven videos ranged from 61% to 86%. The "Welcome" video in Week 1 was viewed for the longest duration (86% [1:02 of 1:12]), whereas the "Goal-Setting" and "Scheduling" videos in Week 3 had the lowest view time percentages (61% [1:44 of 2:50] and 67% [1:01 of 1:31], respectively).

Intervention Acceptability

Weekly Feedback Questionnaires

For each week of the intervention, participants provided positive ratings (i.e., scores above the "neutral" anchor point) on all parameters of acceptability (ratings ranging from 4.81 to 6.69 out of 7). **Figure 1** presents mean ratings of each parameter of acceptability for each week of the intervention. The overall mean acceptability for Weeks 1 through 4 was 5.74 (SD = 0.77), 5.89 (SD = 0.64), 6.19 (SD = 0.34), and 6.02 (SD = 0.50), respectively.

Open-Ended Feedback

Categories, frequencies, and quotes emerging from the content analysis are presented in **Table 3**. Overall, participants indicated that *Plan to Move*: (1) was easy to use and understand; (2) favorably shifted their self-awareness and personal meaning of PA; and (3) provided value and potential for future use pertaining to the learned skills and strategies. Participants' recommendations for improvements related to formatting of the independent activities, including more examples on self-regulation, and providing information about PA guidelines, sample exercises, and accessible facilities and sport opportunities.

Social Cognitive and Behavioral Outcomes

Table 4 summarizes the means, standard deviations, and RM-ANOVAs for all social cognitive and PA behavior outcomes. There were significant and large effect sizes showing increased task ($n^2p = 0.28$) and barrier self-efficacy ($n^2p = 0.24$) between baseline and 1-month post-intervention (adjusted ps = 0.01 and 0.04, respectively). There were significant and large effect

TABLE 2 | Implementation fidelity and intervention compliance outcomes.

Intervention component	**Implementation fidelity**			**Intervention compliance**			
	Dose, *n*	**Number of days to deliver,[a] M (SD) [Range]**	**Adherence: On-time delivery, *n***	**Online session completion, *n***	**Independent activity completion, *n***	**Number of days to complete,[b] M (SD) [Range]**	**On-time completion of each week, *n***
Baseline[c]	16	–	16	–	–	–	–
Overall[d]	100%	4.35 (0.41)	98%	94%	98%	5.28 (4.02)	81%
Week 1	16	–	16	16	16	3.18 (2.32) [0–7]	16
Week 2	16	4.00 (1.93) [0–8]	15	14	16	5.88 (3.59) [0–13]	12
Week 3	16	4.25 (2.08) [0–7]	16	15	15	5.38 (4.15) [0–15]	12
Week 4	16	4.81 (2.34) [0–7]	16	15	16	6.69 (5.03) [1–15]	12
1-week post-intervention[c]	16	6.94 (4.02) [3–15]	15	–	–	–	–
1-month post-intervention[c]	16	22.31 (4.22) [17–30]	14	–	–	–	–

M, mean; SD, standard deviation.

[a] *Number of days between participants' completion of each week of the intervention and delivery of the following week.*

[b] *Number of days for participants to complete each week upon receiving materials (i.e., link to online session and independent activity) via email.*

[c] *Refers to the three assessments conducted by the researcher and do not represent weekly sessions.*

[d] *For outcomes referring to number of days, values reflect an overall average of the mean number of days to deliver and complete each week of the intervention. Percentage values reflect the overall percentage of timely delivery, online session and independent activity completion, and timely completion of each week of Plan to Move out of the 64 total instances of intervention delivery (4 weeks × 16 participants).*

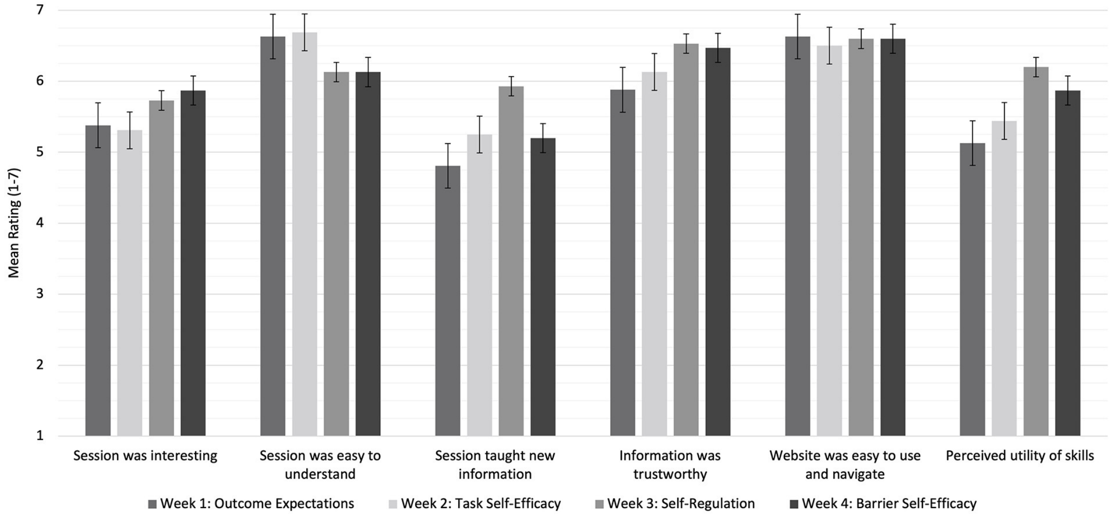

FIGURE 1 | Mean weekly ratings of intervention acceptability parameters.

sizes showing increased goal-setting and planning and scheduling behaviors (n^2ps $= 0.42$) between baseline and 1-week post-intervention (adjusted ps $= 0.01$), and from baseline to 1-month post-intervention (adjusted ps $= 0.01$). Despite a significant main effect, significant *post-hoc* effects were not found for goal-setting self-efficacy (all adjusted ps > 0.05). No significant changes were found in outcome expectations or planning and scheduling self-efficacy. Lastly, there was a significant and large effect size showing increased self-reported PA behavior ($n^2p = 0.26$) between baseline and 1-month post-intervention (adjusted $p = 0.04$).

DISCUSSION

This pilot study explored the feasibility and potential social cognitive and behavioral outcomes of an online SCT-based PA intervention for YPD. Intervention feasibility was supported by: (1) high implementation fidelity, (2) high intervention

TABLE 3 | Open-ended feedback content analysis summary.

Topic	Category	Frequency (% of total)	*n*	Sample quote
Satisfaction	User-friendliness	35 (18%)	16	"I really liked the websites. The websites were working. […] I liked how this one was working good, I just clicked a link and got there. It was easy to use."—Sophie "I thought they were really easy to use because there weren't a lot of different links, and only one button I had to press when I got through one section."—Leah
	Clarity	38 (19%)	16	"One thing I noticed that really nice was the videos. I liked how they summed everything up really nicely. Like from reading the website to watching the video, it made it more understandable."—Logan "[…] there wasn't a lot of information on one page, so it wasn't overwhelming. Each section had the right amount of information. The activities, like the way they were explained, it was broken down really well, and I understood exactly what I had to do. It helped me understand the information you have on the website and put it into context for me."—Leah
Potential impact	Increased self-awareness of their PA	25 (13%)	12	"When you're talking about incorporating physical activity into a busy schedule [*in Week 3*], it really helped a lot because I do have a busy schedule. Now I can find key points where I can fit physical activity here or there or whenever. It changed how I looked at things and how much time I have."—Grace "When there's a day when I realize maybe I'm doing too much, I can schedule physical activity for other days when I'm not doing as much. Especially with the chart [a self-monitoring tool provided in Week 3], it was really easy to see where I could plan my physical activity."—Molly
	Positive reframing of PA	13 (7%)	7	"I think what I realized the most was that physical activity is not just going to the gym or playing a certain sport. Like there's a lot of things you can do in your daily life that can count for physical activity, like into your daily routine without having to disrupt it. Like taking longer walks. After this program, I took up one new sports activity. I got inspired to learn [something new], so I started taking ice skating classes. It's really fun—it's challenging but I'm just going at a slow pace and I'm having fun."—Amanda "By only doing 10 min at a time, it makes it more manageable and less intimidating."—Charlotte
Utility	Value of learned skills	24 (12%)	11	"A big thing for me is not having time. These skills help me understand how to plan to have more time to be active. Also, this program reinforced the idea of regular goal-setting and how it can help me get more physical activity."—Ethan "Scheduling helped me see when I had free time. I knew I could use that time to do physical activity. It made sure I wasn't sitting around and wasting time."—Zara
	Current and future use of learned skills and strategies	30 (15%)	14	"I think I'd use the things I learned to continue working out. Like the reminders definitely are a huge help and have been something I've been using since I learned about them. Instead of just putting it in my calendar and forgetting to do it, putting it in my calendar and setting reminders really helped me remember to actually go and achieve that goal or workout that I wanted to do."—Camila "I liked having a set plan and sticking to that plan. I put reminders on my phone to stretch during homework breaks, or while watching TV. I liked it, because I need to do those stretches for my spasms anyway."—Chloe
Recommended improvements	Formatting	15 (8%)	13	"I found the first few were properly formatted, […] but there were a few pictures covering the questions. Maybe have like on the website that you used […], have it on the website so then you don't have the document where the formatting gets messed up."—Logan "I was having some issues with formatting. I would not recommend doing the activities on Word. If there was like an online program that would work universally, that would probably be better."—Olivia
	Additional information	15 (8%)	7	"I would like to know more about nutrition and stretching and all that kind of stuff after physical activity, like how to take care of your body if you're sore. How often you should be exercising, as in like when you should take a rest, and rotating muscle groups, would have been really helpful. […] This program may be good for teenagers or like younger teens who haven't yet been educated on the benefits of physical activity. I think for me, it may be really nice if the program had links to sport associations to get you involved or accessible facilities to stay fit. Sample exercises, that kind of stuff."—Olivia "You could give us links to gyms where we could go or something like that. Maybe accessible places where we can go workout. It's not always helpful to just tell us about the tools to get active, but you have to kind of reinforce that with where we can get active."—Elliot "I think like when you're talking about goal-setting, there were some really good examples, but I just feel like maybe if you were like a little more descriptive it would be better."—Grace "[…] providing more scientific information for some things in Week 1 [*referring to outcome expectations],* like some studies or background. You see the value for those things. That would also help with motivation and drive to start being active."—Logan

All participants were assigned pseudonyms to protect their anonymity.

compliance, (3) positive ratings on indicators of acceptability, and (4) participants' perceived satisfaction, impact, and utility of the intervention. Participants experienced significant and large-sized increases in task and barrier self-efficacy, goal-setting, planning and scheduling, and self-reported PA. These findings are encouraging and demonstrate that an SCT-based online PA

TABLE 4 | Means, standard deviations, and one-way RM-ANOVAs for social cognitive outcomes and self-reported PA.

Variable [Potential score range]^a	Baseline, M (SD) [Actual range]	1-week post, M (SD) [Actual range]	1-month post, M (SD) [Actual range]	*F*-ratio (df)	*p*	η^2p
1. Outcome expectations [22–396]	264.69 (61.44) [164–396]	290.56 (65.75) [168–396]	281.50 (61.08) [163–396]	2.66 (2, 11)	0.09	0.15
2. Task self-efficacy [1–7]	3.43 (0.61) [2.29–4.57]	3.79 (0.45) [2.86–4.29]	3.81 (0.59) [2.57–4.57]	5.89 (2, 11)	0.01	0.28
3. Goal-setting self-efficacy [0–100%]	69.84 (14.56) [45.00–95.00]	77.47 (15.85) [51.25–100.00]	75.70 (11.39) [57.50–95.00]	4.22 (2, 11)	0.02	0.22
4. Planning and scheduling self-efficacy [0–100%]	72.19 (17.05) [32.14–96.43]	75.96 (16.54) [38.57–98.57]	75.89 (14.13) [40.00–92.14]	1.79 (2, 11)	0.20	0.11
5. Barrier self-efficacy [1–7]	4.29 (0.90) [2.75–5.88]	4.83 (1.14) [2.63–6.50]	4.81 (1.08) [3.38–6.88]	4.66 (2, 11)	0.02	0.24
6. Goal-setting behavior [10–50]	28.69 (7.27) [17–39]	34.44 (8.49) [18–46]	35.31 (6.06) [19–44]	11.01 (2, 11)	<0.001	0.42
7. Planning and scheduling behavior [10–50]	28.44 (8.64) [16–45]	34.38 (8.02) [19–49]	33.88 (6.49) [20–45]	10.66 (2, 11)	<0.001	0.42
8. Self-reported weekly minutes of PA	248.13 (171.34) [50–590]	320.75 (184.64) [40–670]	415.94 (365.46) [45–1,185]	5.32 (2, 11)	0.02	0.26

^{a}*higher scores reflect improved outcomes for each variable.*
M, mean; SD, standard deviation.
η^2p, partial eta squared, such that 0.01 = small, 0.06 = medium, and 0.14 = large effect sizes (25).

intervention for YPD is feasible and may yield positive social cognitive and behavioral change over a short period.

Given their novelty, the feasibility of online PA interventions for YPD was largely unknown. This study contributes to the generation of knowledge on related feasibility outcomes and can inform the procedure of future online PA interventions for YPD. Notably, nearly perfect implementation fidelity was achieved, demonstrating that the delivery of an online PA intervention as intended in YPD is practical. The digital and self-led nature of the intervention reduced the magnitude of the facilitator's involvement substantially in comparison to the degree of involvement that could be expected in traditional in-person or facilitator-led interventions. This reduced level of facilitator involvement, and potential burden, was likely a contributing factor to the high implementation fidelity observed in the current study. As such, these findings provide further support for the value of leveraging technology in the delivery of PA interventions for YPD.

Considering the challenges related to retention in technology-based interventions (12), high intervention compliance (>90%) and relatively low attrition (20%) in the current study is promising. Text message reminders (11) and the short duration of the intervention (27) may have facilitated greater compliance and retention than a longer intervention would have. In addition, the self-led nature of the intervention may have allowed participants some degree of flexibility in comparison to a traditional in-person or facilitator-led intervention, where scheduling or other constraints may lead to poor compliance or attrition. In contrast, in the current intervention, participants were given a certain degree of autonomy to complete each week of the intervention (i.e., within 7 days). Further, given the frequent use of YouTube$^{\text{TM}}$ by YPD (28), embedding YouTube$^{\text{TM}}$ videos may have offered a salient method of communication that encouraged some degree of continued engagement. The use of YouTube$^{\text{TM}}$ videos was novel, and thus, expected outcomes relating to its feasibility were unknown. Despite 69% average viewership of the total minutes of the embedded videos, participants' positive response to the YouTube$^{\text{TM}}$ videos, as demonstrated through their open-ended feedback during the 1-month post-intervention assessment, suggests that it may be worthwhile to incorporate YouTube$^{\text{TM}}$ videos in future PA interventions for YPD. Further work is needed though to determine the appropriate video length and content (e.g., knowledge vs. examples demonstrating the application of skills) to optimize YPD's sustained engagement and exposure to intervention content.

With regard to intervention acceptability, participants provided positive ratings (i.e., scores above the Likert scale's "neutral" anchor point) on all indicators of acceptability in the weekly feedback questionnaires. Overall, participants indicated that each week of *Plan to Move* was easy to navigate, easy to understand, and provided credible information that they would likely use in the future to manage their PA behavior. Notably, participants' ratings of the perceived novelty and utility of learned skills was highest in Week 3, which targeted self-regulation. Similar intervention acceptability outcomes were revealed through participants' open-ended feedback provided during the 1-month post-intervention assessment. Participants shared that, overall, they were satisfied with *Plan to Move*, largely as a result of the intervention's user-friendliness (e.g., simple navigation) and clarity. Furthermore, participants indicated that the learned self-regulation skills would likely help them manage their PA behavior in the future. Overall, participants' open-ended feedback aligned with the findings from the weekly feedback questionnaires.

Despite their overall perceived acceptability of *Plan to Move*, participants also provided valuable recommendations on how to improve future content and design. In particular, older participants (aged 20–21 years) expressed interest in learning about PA guidelines, sample exercises, and resources on accessible facilities and sport opportunities. Although PA prescription was not within the scope of the intervention, these suggestions provide insight on the type of information older youth may be seeking when participating in a PA intervention. Participants also indicated a preference for the independent activities to be embedded within the online sessions rather than as an offline Word document to mitigate challenges related to formatting incompatibilities across different operating systems and versions of software. Thus, future PA interventions for YPD should streamline all components of the intervention within one interface to deliver a more integrated experience with less potential for formatting incompatibilities.

Participants' enhanced social cognitions was a positive outcome. Contrary to previous evidence (11), task self-efficacy increased. This discrepancy may be explained by the provision of various self-regulatory strategies in the current intervention. Emphasizing self-regulation may have counteracted potential negative effects on self-efficacy by providing participants with a set of tools to manage salient PA-related challenges that were potentially heightened by participating in the intervention itself. From a theoretical perspective, self-efficacy is a direct determinant of health behavior and also has indirect effects on behavior through intermediate determinants (e.g., self-regulation). Thus, future PA interventions should target self-efficacy *and* self-regulation to maximize potential for behavior change. The observed increase in barrier self-efficacy is consistent with previous evidence (10) and should continue to be targeted.

Although goal-setting and planning and scheduling behaviors increased, this was not complemented by an increased self-efficacy to engage in those behaviors. This observation warrants consideration of the role of parents of YPD. Given the unique challenges that YPD experience, parents are a vital source of support and often manage their child's schedule and act as a prompt to execute plans (29). Thus, YPD may not feel confident in their ability to self-manage goals. Shifting the responsibility of self-regulation from parent to child can enhance independence and better prepare YPD to self-manage their PA. Thus, self-regulation should be targeted in such a way that also enhances YPD's self-efficacy to engage in self-regulation behaviors.

Contrary to previous evidence (10), outcome expectations did not increase. Participants' open-ended feedback suggests that outcome expectations may need to be targeted differently in YPD. For example, participants expressed interest in learning about the scientific literature supporting the benefits of PA. This approach may substantiate the benefits of PA and be more effective than listing benefits that YPD are likely aware of. Future work should explore how outcome expectations can be more effectively targeted and enhanced in YPD, as SCT constructs are reciprocally interrelated and have direct effects on behavior (9).

Increased self-reported PA between baseline and 1-month post-intervention was an unexpected but welcomed outcome, as previous evidence demonstrated no significant increases in self-reported or objectively measured PA following intervention in YPD (11). Targeting known theoretical correlates of PA for youth in the current study may have facilitated an increase in PA. Despite the LTPAQ-SCI being a validated measure of PA in persons with physical disabilities (24), participant knowledge of the intervention's objective to enhance PA may have caused response bias and warrants caution in the interpretation of this observed increase. Although utilizing wearable devices (e.g., accelerometer) for the measurement of PA would counter such bias, this approach would pose challenges in the reliable measurement of PA in non-ambulatory YPD (30).

This was the first study to evaluate the feasibility and outcomes of an online theory-based PA intervention in a diverse sample of YPD. Focusing on the end-user in the current study allowed for an understanding of what elements of the intervention did and did not work from a usability and feasibility perspective. Further, the use of theory allowed for insight on constructs that were enhanced and others that may need to be targeted differently in YPD (i.e., outcome expectations and self-regulatory efficacy).

Despite these strengths, the lack of a control group precludes the determination of whether the observed changes would or would not have occurred in the absence of an intervention. Thus, the observed changes in participants' social cognitive and behavioral outcomes are not an indication of intervention efficacy nor can they be attributed as an outcome of the intervention itself. Furthermore, although discussion topics were introduced neutrally and participants were unaware of who developed the intervention, there was potential for bias in participants' open-ended feedback, as the discussions were conducted by the researcher delivering the intervention. Future implementation at a larger scale should be appropriately powered and include a control group and longer follow-up period to minimize sampling bias, enhance generalisability, determine efficacy, and elucidate longer-term outcomes. In consideration of implementation at a larger scale, although the use of technology in the current study mitigated the environmental barriers that may otherwise preclude YPD from their in-person participation in PA interventions, it is important to acknowledge that this intervention delivery approach can pose an alternative set of barriers related to inequitable access to technology (e.g., computer, Internet, and software licenses). Although access to technology did not pose any challenges in the current study, strategies to address these potential barriers in the emergence of progressively technology-based intervention approaches must be considered to manage social inequities and deliver a comprehensive PA promotion strategy to YPD that minimizes the impact of a spectrum of barriers and does not drive further health inequities.

Findings from this study support feasibility and can guide the development and implementation of future online PA interventions for YPD. Participants' enhanced social cognitive and behavioral outcomes demonstrates the potential benefit that YPD may experience from participating in an intervention of this nature. Continued research on the topic of online theory-based PA interventions is critical for creating high-quality opportunities for YPD to learn strategies that enable them to enhance and self-manage their PA and overall health. These benefits may not otherwise be accessible to YPD without this alternative method of intervention delivery, which should therefore be considered in the development of future PA promotion strategies for this population.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Health Sciences Research Ethics Board at the University of Toronto. Verbal informed parental consent and verbal youth assent were provided for participants under the age of 18 to participate in this study, and participants over the age of 18 provided verbal informed consent. Verbal informed consent and assent were documented.

AUTHOR CONTRIBUTIONS

RS designed the study with input from AEL-C, JC, and KPA-N. RS developed and delivered the Plan to Move intervention, recruited participants, collected and analyzed data, and lead the writing of the manuscript. KPA-N supervised the execution of the study. All authors contributed to the review, editing, and final approval of the manuscript.

FUNDING

RS received funding from the Social Sciences and Humanities Research Council of Canada—Canada Graduate Scholarships to conduct this research. Additional funds from the Social Sciences and Humanities Research Council of Canada (Grant #895-2013-1021) supported the conduct and publication of this study.

ACKNOWLEDGMENTS

The authors acknowledge the contributions of the participants and their families and extend their gratitude to the community partners and organizations who provided support during recruitment.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fresc.2021.651688/full#supplementary-material

Appendix A | Content overview of weekly sessions, YouTube^{TM} videos, and independent activities of *Plan to Move*.

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Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers in Rehabilitation

BRIEF RESEARCH REPORT
published: 07 September 2021
doi: 10.3389/fresc.2021.690046

Characterizing Cycling Smoothness and Rhythm in Children With and Without Cerebral Palsy

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Stationary cycling is a practical exercise modality in children with cerebral palsy (CP) that lack the strength for upright exercises. However, there is a lack of robust, sensitive metrics that can quantitatively assess the motor control during cycling. The purpose of this brief report was to characterize the differences in motor control of cycling in children with CP and with typical development by developing novel metrics to quantify cycling smoothness and rhythm. Thirty one children with spastic diplegic CP and 10 children with typical development cycled on a stationary cycle. Cycling smoothness was measured by cross-correlating the crank angle with an ideal cycling pattern generated from participant-specific cadence and cycling duration. Cycling rhythmicity was assessed by evaluating the revolution-to-revolution variability in the time required to complete a revolution. Statistically significant differences ($p < 0.001$) using the Wilcoxon Rank Sum test were found between the two groups for both the metrics. Additionally, decision tree analysis revealed thresholds of smoothness <0.01 and rhythm <0.089–0.115 s for discriminating a less smooth, irregular cycling pattern characteristic of CP from typical cycling. In summary, the objective measures developed in this study indicate significantly less smoothness and rhythm of cycling in children with CP compared to children with typical development, suggestive of altered coordination and poor motor control. Such quantitative assessments of cycling motion in children with CP provide insights into neuromotor deficits that prevent them from cycling at intensities required for aerobic benefits and for participating in cycling related physical activities with their peers.

Keywords: recumbent cycling, rehabilitation, physical activity, motor control, fitness

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Specialty section:
This article was submitted to Disability, Rehabilitation, and Inclusion, a section of the journal Frontiers in Rehabilitation Sciences

***Received:** 02 April 2021*
***Accepted:** 09 June 2021*
***Published:** 07 September 2021*

Citation:
Sansare A, Behboodi A, Johnston TE, Bodt B and Lee SCK (2021) Characterizing Cycling Smoothness and Rhythm in Children With and Without Cerebral Palsy. Front. Rehabilit. Sci. 2:690046. doi: 10.3389/fresc.2021.690046

INTRODUCTION

Cerebral Palsy (CP) is a neurodevelopmental disorder of movement and posture that results from an injury to the fetal or infant brain (1). Children with CP typically present with motor deficits such as altered muscle tone and muscle weakness, and may experience impaired sensory and cognitive impairments (2, 3). Although CP itself is a nonprogressive disorder of the brain, the impairments and functional limitations associated with CP are progressive, with many children becoming less independent with functional mobility as they enter their teenage years (4–6).

Children and adolescents with CP participate in less habitual physical activity and are sedentary for more than twice the maximum recommended amount (7, 8). Unfortunately, many children with disabilities are unable to meet global physical activity recommendations due to functional impairments that limit the type of exercise activity they can participate in (6) as well as they may be limited from safely performing exercise or accessing the equipment needed to do so (9, 10). The disparity is often exacerbated by the interventions used to abate musculoskeletal and soft tissue changes that contribute to deformity, muscle tightness, and joint contractures. Selective dorsal rhizotomies, muscle/tendon lengthening procedures, serial casting, botulinum toxin injections, corrective bony procedures and the like, further compromise muscle strength by removing spasticity thereby unmasking muscle weakness, by putting muscles at unfavorable lengths for force generation, and by forced periods of prolonged immobility required by the corrective procedures (11–15). Thus, as children with CP mature, they have marked difficulties in maintaining fitness and functional ability. Hence, it is critical to develop exercise modalities that enable children with CP with limited or marginal ambulatory abilities to safely engage in physical activities.

Recumbent stationary cycling has been proposed as a safe, enjoyable, and practical exercise modality for children with CP that lack the postural control and strength necessary for upright exercises (16–18). Individuals with CP, however, are known to have impairments such as agonist-antagonist co-contraction and abnormal muscle tone (19), which may lead to irregular, halted progression of revolutions during cycling (20), thus affecting the rhythmicity and smoothness of cycling. Cycling with poor smoothness, e.g., arrested progression of revolutions and poor rhythmicity may result in inefficient cycling and reduced intensity of the exercise, and thereby, lead to reduced efficacy. Cycling with maladaptation will further lead to reinforcement of atypical movement patterns. Thus, it is critical to evaluate the motor control of cycling to train correct neuromuscular strategies for more optimal benefits from cycling. Although cycling performance has been previously evaluated in terms of muscle activation, kinematics and kinetics (19, 21) there are no studies that quantitatively describe motor control during cycling.

Smooth and rhythmic movements are a characteristic of well-developed motor control (22). While several smoothness metrics based on upper limb reaching movements, such as jerk (the time derivative of acceleration) and spectral analysis, have been proposed (23), they are affected to different degrees by measurement noise, movement duration, and periods of movement arrest. Using these metrics for detecting differences in smoothness during upper limb motion between healthy controls and individuals with stroke, cerebellar disorders and Parkinson's disease has led to mixed results (24). Such metrics are especially problematic in CP for a couple of reasons. First, taking higher order derivatives of abrupt, jerky movements that are characteristic in individuals with CP leads to outputs that are closer to the metric's ceiling values. This can result in reduced sensitivity of the measure during within- and between-participant comparisons. Second, most smoothness metrics do not quantify the temporal aspect of motion, such as regularity and variability in duration of cycling revolutions, which are important components of motor control. Thus, there is a need for robust, dimensionless, and sensitive measures for evaluating smoothness and rhythm of cycling in CP. Such metrics of cycling smoothness and rhythm may enable more effective corrective training strategies that could make cycling exercise more widely adapted by individuals with CP. With further rigorous testing on sufficient sample sizes, such metrics can have the potential to serve as tools to track changes in motor impairments in CP and the effect of treatments, such as functional electrical stimulation (FES) and biofeedback-augmented cycling, on improving motor control. The aim of this study is to characterize differences in motor control of cycling in children with CP and with typical development (TD) by developing novel metrics to quantitatively describe cycling smoothness and rhythm. We hypothesize that children with CP will demonstrate less smoothness and rhythm of cycling motion compared to those with typical development.

METHODS

Children with spastic diplegic CP were recruited through the outpatient CP clinic at Shriners Hospital for Children, Philadelphia and local referral sources. Appropriate Institutional Review Board, administrative permissions were obtained. Additionally, written informed consent from the parent/guardian of the participants and written assent from the participants were obtained. The data from children with TD was obtained from a pre-existing dataset of 10 healthy, typically developing children recruited in a hospital setting through advertisement at the hospitals, local community-based sources, siblings of previous participants, and word of mouth. None of the children with TD were patients at the hospital. All participants were screened by a physical therapist for the inclusion and exclusion criteria (**Table 1**).

The system used for the CP group consisted of a commercially available recumbent sport tricycle (www.kmxkarts.co.uk) fitted with shank guide orthoses to control for excess hip adduction and abduction movement (**Appendix A**) (25). The bicycle crank and spindle assembly was instrumented with sensors to indicate crank position and cadence. The cycling assessment system for the children with TD consisted of a semi-recumbent, free-standing Restorative Therapies, Inc. bicycle (Baltimore, MD) attached to a therapy bench. The children in the CP group were all novice cyclers, and hence performed 20-min practice sessions twice daily for 3 days before the assessment while the children with TD performed a 10 min practice session. All children were allowed rest breaks as needed during the practice sessions. During the assessment, the children in the CP group cycled for an average of 30 ± 13 s (mean ± SD) while children with TD cycled for 15–30 s. Additionally, children with TD were asked to cycle at a target cadence of 60 rpm. However, the participants in CP group had difficulties in attaining the 60 rpm target cadence. Hence, they were all encouraged to pedal as fast as they could to get cycling as close to 60 rpm as possible. The ergometer resistance was calculated using the same formula in both CP and TD groups and was adapted from Doré et al. (26). Load (in newton-meters)

TABLE 1 | Inclusion and exclusion criteria.

Inclusion criteria	Exclusion criteria
• Ages 10–18[a] • Diagnosis of spastic diplegic CP[b] • GMFCS II, III, or IV[b] • Adequate range of motion of the hips, knees, and ankles to allow pedaling • Visuoperceptual skills and cognitive/communication skills to follow multiple step commands for attending to exercise and data collection • Ability to communicate pain or discomfort with testing and training procedures	• Lower-extremity orthopedic surgery or traumatic fracture within the past 6 months • Lower-extremity joint pain during cycling • Spinal fusion extending to the pelvis • Hip, knee, or ankle joint instability or dislocation • Lower-limb stress fractures in the past year • Symptomatic or current diagnosis of cardiac disease as assessed by the American Heart Association guidelines for cardiac history • Current pulmonary disease or asthma and taking oral steroids or hospitalized for an acute episode in the past 6 months • Severe spasticity in legs (score of 4 on the Modified Ashworth Scale)[b] • Severely limited joint range of motion or irreversible muscle contractures that prevented safe positioning on the cycle[b] • Diagnosis of athetoid or ataxic CP [b]

[a] *Age range for participants with typical development was 13–19 years.*
[b] *Participants with cerebral palsy (CP) only.*

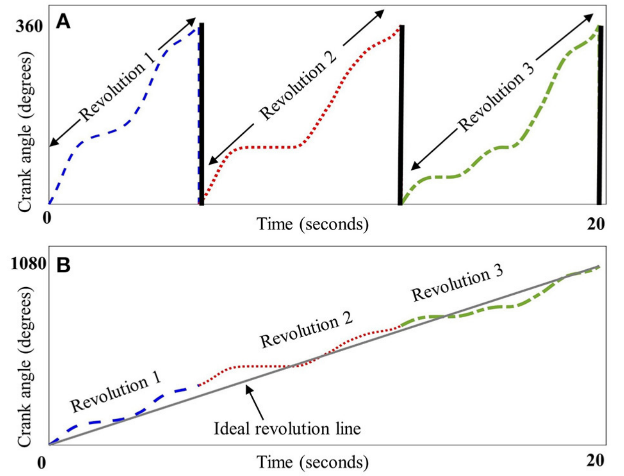

FIGURE 1 | Schematic representation of crank angle smoothness. Panel **(A)** depicts crank angle (plotted against time in seconds) for three representative revolutions from a child with CP, each dashed section depicting one revolution from 0° to 360°, thick black lines indicate the discontinuity between 360° and 0° at the end of each revolution. Panel **(B)** depicts the concatenation of these revolutions, resulting in a linear form that was cross-correlated with a line depicting an ideal, smooth revolution (straight gray line).

= 0.49 N/kg × body weight (in kilograms) × crank arm length (in meters). The CP (R01HD062588) and TD datasets (19) were from two separate larger studies. Despite the different systems for children with CP and TD, the overall set-up was custom adjusted according to the same specifications for each participant based on their anthropometric data (**Appendix A**). Because the same standardized system set-up, including crank arm length, seat-to-pedal distance and seat-to-greater trochanter distance, were used for both the groups, we do not expect the different cycling systems to contribute appreciably to the between-group differences that may be observed. Data were analyzed using customized software (MatLab, The Mathworks, Inc.) and statistical software (JMP®, Version 14.3.0, SAS Institute Inc.).

Data Analysis

Crank angle data were lowpass filtered at 5 Hz and plotted against time, the result being a sawtooth waveform indicating the angle of the recumbent cycle's crank as the trial progressed. To eliminate potential pedal acceleration and deceleration influences, the first and the last revolution of the crank were discarded. As crank angle data are circular, there is a discontinuity every time the angle value crosses from 360°→0° (**Figure 1A**). To eliminate this discontinuity, crank angle was converted from repeating 0–360° epochs to a linear form by concatenating the angle data and appending them in series. The resultant angle-in-series data was a time series representing the angular progression of crank from zero to 360 × the number of revolutions (**Figure 1B**). To quantify the deviation of each participant's angle-in-series from the smoothest possible crank angle, the angle-in-series was cross-correlated with a straight line that connected the beginning to the end of angle-in-series' data points. This straight line, considered the participant -specific ideal crank angle, represented the smoothest transition from 0°→360°. The duration of this ideal straight line for each cycling trial was the same as the cycling duration of the observed pattern to account for possible influences of the cycling speed and duration on smoothness. Also, to eliminate any influence of cadence on smoothness, the number of revolutions in the ideal pattern were the same as that in the observed cycling trial. Thus, an ideal cycling pattern was "custom-made" for each participant based on their own speed, cycling duration and cadence.The calculation of the cross-correlation between angle-in-series and ideal crank angle for the time lag *n*, including the formula used to calculate it, is further described in **Appendix B**. The maximum of the cross-correlation of the angle-in-series and ideal crank angle was then normalized to the maximum of ideal line's autocorrelation, which is the cross-correlation of the signal with itself, to make it dimensionless for better comparison. The results were expressed as the smoothness measure. Higher values indicate less smooth cycling motion.

To quantify the temporal characteristics of cycling, or in other words, to assess how rhythmic and regular the cycling pattern was, the variability of the time taken for completing each revolution in a cycling trial was measured by computing its standard deviation. Therefore, similar to the definition of gait rhythmicity as stride-to-stride variability in gait timing (27, 28), we defined cycling rhythmicity as revolution-to-revolution variability in the time required to complete a revolution. Thus, the higher the standard deviation, the higher the variability and lower the rhythmicity of each revolution.

We analyzed between-group differences by performing a nonparametric Wilcoxon Rank Sum test. To further support the ability of smoothness and rhythm metrics to discriminate between the typical cycling pattern and a less smooth, irregular pattern seen in CP, we performed a decision tree analysis using the Partition routine within JMP using default settings. The ability of a decision tree to accurately classify group membership is enhanced by individual measures with distributions immediately distinguishable between groups and can be refined further with additional measures that explain group membership conditional on earlier branches in the tree. Decision trees were built separately, using smoothness or rhythm for the initial branch split, to determine threshold values to distinguish between CP and TD cycling patterns, and then refined if possible by the remaining predictor. In cross validation, validation sets were randomly formed with ∼80% of the data used for training the algorithm and establishing the decision rules and the remaining ∼20% used as a validation set on which the rules could be applied. We replicated the process three times to probe the sensitivity of the fit to the random allocation of training and validation. An additional probe of sensitivity was conducted using the JMP software implementation of 5-fold cross validation, which we also ran three times for each predictor to build confidence in the approach through the generalized R^2 reported. Confusion matrices report the number of correct and incorrect predictions for CP/TD cycling pattern using decision tree-derived thresholds for smoothness and rhythm metrics.

Lastly, to explore the sensitivity of our metrics to aberrant revolutions, we performed simulation analysis using custom MATLAB software. We generated alternate datasets from the original dataset in the following way:

1. To explore how a single aberrant revolution affects smoothness, we removed the most aberrant cycle in terms of smoothness, i.e., the most unsmooth revolution from each participant's trial. Thus, we generated an alternate dataset from the original dataset without the most unsmooth revolution.
2. To explore how a single aberrant revolution affects rhythm, we repeated the same process for rhythm, where we generated an alternate dataset without the revolution with worst rhythmicity for each participant.
3. To investigate how the order in which the aberrant revolution occurred in a trial affects smoothness, we generated an alternate dataset by shuffling the positions of the revolutions in a trial.

Next, we recalculated the smoothness and rhythm values for the alternate datasets mentioned above. The difference between the two datasets was analyzed using paired *t*-tests, where the original and alternate values for each participant formed a single pair. Because shuffling the revolutions would not change the variability of the revolutions and in turn would not change the rhythm values, no further statistical analysis was performed for rhythm for the third scenario listed above.

RESULTS

Thirty-one ambulatory adolescents with CP were recruited, with Gross Motor Function Classification System (GMFCS) levels II–IV (level II, III, and IV had 10, 10, and 11 participants, respectively). There were six females in the CP group and seven females in the TD group. There were no significant between-group differences for age ($p = 0.127$) and BMI ($p = 0.570$). The mean [standard deviation (SD)] age was 13.7 (2.6) years for children with CP and 14.9 (1.4) years for children with TD. The mean (SD) BMI was 20.3 (5.5) kg/m^2 for children with CP and 22.6 (5.4) kg/m^2 for children with TD. By inspection, **Figure 2** boxplots reveal that the distributions for smoothness and rhythm each appear different for children with CP and TD (**Figure 2**). Extreme observations or outliers were cross-checked through visual inspection of the raw data and visualization of the crank angle against time, which revealed that these were valid measurements and not measurement errors. The two-sided tests yielded normal approximation z-values of −3.81 (Smoothness) and −4.69 (Rhythm), each statistically significant ($p < 0.001$). The mean smoothness and rhythm (mean ± standard error) for children with CP [0.039 ± 0.010 (dimensionless) and 1.672 ± 0.583 (s), respectively] were significantly higher than that for children with TD (0.006 ± 0.001 (dimensionless) and 0.005 ± 0.001 (s) respectively). Higher values for both metrics indicate less smoothness and less rhythmicity of cycling motion.

We explored the potential of our measures to accurately discriminate the cycling pattern as being that of a child with CP or TD. Once either smoothness or rhythm was included in the decision tree analysis model, the second metric added no additional predictive advantage, resulting in a single decision rule for each metric. The decision rule for smoothness revealed smoothness >0.01 as threshold for predicting cycling pattern characteristic of the CP group for all validation sets. The decision rule for rhythm revealed rhythm >0.115 s as threshold for predicting a CP cycling pattern for validation set 1 and >0.089 s for validation sets 2 and 3. Additionally, a software generated five-fold cross validation on the same data yielded a generalized $R^2 = 0.99$ in each of the three runs. The details about the training and validation confusion matrices for the decision tree are depicted in **Appendix C**.

Our exploration of the sensitivity of the metrics showed that there were no significant differences between the original smoothness values and the values generated after removing the most unsmooth revolution ($t = -0.287$, df [40], and $p = 0.776$). There were, however, significant differences between the original rhythm values and the values generated after removing the revolution with the worst rhythmicity ($t = 2.594$, df [40], and $p = 0.013$). On repeating the same analysis after excluding the participants whose trial had <12 revolutions, there were no significant differences between the original rhythm and the rhythm without the most aberrant cycle ($t = 1.580$, df [19], and $p = 0.065$). Lastly, the shuffling of revolutions did not yield smoothness values that are statistically significant from each ($t = 1.072$, df [40], and $p = 0.145$).

DISCUSSION

The purpose of this study was to develop objective measures to quantify motor control during cycling in children with CP and with TD. We developed two measures, one to assess the quality

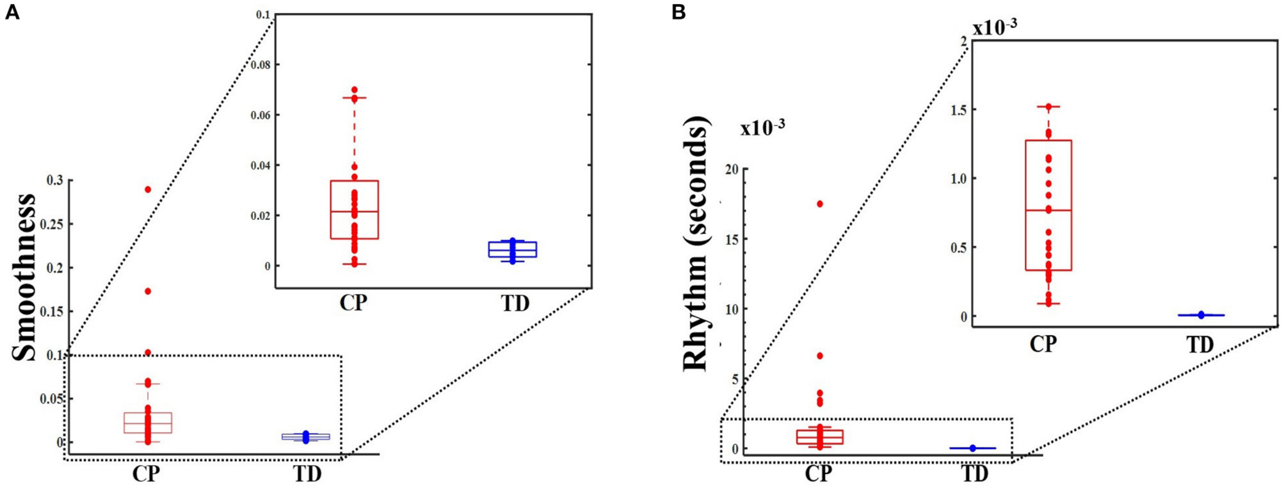

FIGURE 2 | Box and whisker plots, with scattered values (dots) indicating each data point, show increased values, i.e., less smoothness (dimensionless metric) in panel **(A)** (top panel) and for rhythm (in seconds) in panel **(B)** (bottom panel) for CP group (on the left, in red) compared to typically developing (TD) group (on the right, in blue). Higher smoothness values imply a less smooth cycling pattern. Higher rhythm values imply increased variance, and hence, poorer rhythm. The boxes depict the 25th–75th quartile and the horizontal line depicts the median. Inset: Smoothness and rhythm values for majority of the data (excluding the extreme values).

of cycling motion i.e., smoothness, and the second to assess the regularity in the timing of cycling, i.e., rhythm of cycling motion.

Differences Between CP and TD Cycling

Our results show that children with CP cycled with significantly less smoothness as compared to children with TD (**Figures 3A,B**). Thus, the progression of crank angle from 0° to 360° was significantly more halted and abrupt in children with CP. Also, children with CP cycled with significantly less rhythmicity compared to children with TD, i.e., the time taken to complete a cycling revolution was extremely variable in the CP group, leading to irregularity and poor rhythmicity of the motion (**Figures 3C,D**). Thus, both metrics were able to quantify the difference in motor control of cycling between children with CP and TD. These differences may be due to agonist-antagonist co-contraction, increased duration of muscle activation and altered motor strategies previously reported in children with CP during cycling (19, 21). Our results are also consistent with reports of reduced smoothness during upper limb reaching in CP (29) and with video analysis that showed irregular time periods spent within different quadrants of the pedaling cycle (20). Our results collectively with these studies are indicative of altered motor control in CP.

Additionally, the decision tree results further support the ability of the two outcome measures to successfully discriminate between a typical cycling pattern and an abnormal, less smooth, and arrhythmic cycling pattern seen in CP. The decision tree analysis identified empirically derived thresholds for these measures. Smoothness above 0.01 was attributed to the CP group while smoothness below 0.01 was attributed to the typical cycling pattern. Similarly, rhythm scores above 0.089 and 0.115 s distinguished a CP cycling pattern from TD. Obtaining two threshold values from two different training sets for rhythm is not unusual, given the small data set with high variability in the CP group which comprised individuals with different functional capabilities (GMFCS levels II–IV). However, the results of the rhythm confusion matrices are encouraging (**Appendix C**).

Sensitivity of the Metrics

Exploration of the sensitivity of the metrics revealed that the smoothness values were largely unaffected by a single aberrant cycle, implying that while the metric can consistently discriminate between a smooth and unsmooth cycling pattern, it is less likely to be influenced by a single aberrant revolution or an outlier. The rhythm metric significantly changed due to the removal of the most aberrant cycle, implying that it is extremely sensitive to even a single aberrant revolution. However, when the participants with <12 revolutions were excluded from the analysis, a single aberrant revolution was less likely to affect its value. Thus, rhythm is especially more sensitive to deviations caused by single outlier in the absence of sufficient number of cycling revolutions. We caution against using twelve revolutions as an absolute threshold or rule of thumb for collecting the minimum number of revolutions, rather our intent was to demonstrate that too few cycling revolutions might magnify the effect of single aberration on the metric. While another approach to characterizing variability, such as using the coefficient of variation, which is standard deviation divided by mean, may be used to quantify rhythm, it may mask the raw variability that the standard deviation captures. As both standard deviation and mean may simultaneously increase or decrease, the resultant coefficient of variation may remain the same, masking potential pre- to post-intervention changes for a patient. Finally, shuffling of the revolutions in a trial did not affect either metric, implying that the metrics are not affected by the location of the aberrant revolution.

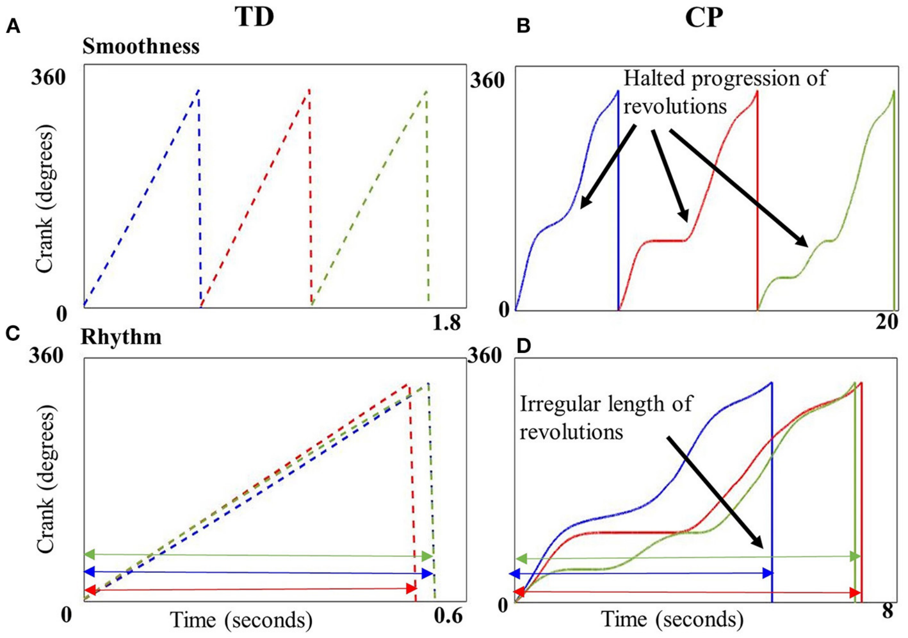

FIGURE 3 | Crank angle plotted against time (seconds) for three example revolutions (blue, red, and green) from a single representative child with typical development (TD) **(A,C)** and CP **(B,D)**. Top panels depict the smooth transition from 0 to 360° in a child with TD **(A)** vs. the uneven, halted progression in a child with CP **(B)**. Cycling revolutions are superimposed on each other in the bottom panels to depict rhythmicity, which shows the consistent length of the revolutions in a child with TD **(C)**, implying better rhythmicity in contrast to the inconsistent length of revolutions in a child with CP **(D)** implying poor rhythmicity.

Existing smoothness metrics, which are especially sensitive to signal-to-noise ratios, result in different smoothness values for the same movement pattern with changes in movement speed. This is because slower movements have lower SNR (signal to noise ratio) than faster movements. Thus, if the smoothness measure is extremely sensitive to changes in SNR, then one would get different results for smoothness of the same cycling pattern at different speeds. The strength of our smoothness metric lies in comparing the observed cycling motion with a "custom-made" participant-specific ideal cycling pattern derived from their own cycling speed, duration, and cadence, thus making possible comparisons across individuals with different instantaneous speeds and cadences. This attribute is especially important while assessing motion in a clinically heterogeneous disorder such as CP, where individual may vary vastly in their functional abilities, leading to different cycling speeds and durations. Our smoothness metric, in essence, enables the evaluation of motor control of the cycling motion, irrespective of the cycling speed and cadence.

Clinical Application for Enhancing Physical Activity

Quantitative assessment of motor control during cycling may provide insights into some of the potential impairments, such as poor rhythmicity and halted unsmooth motion that may hinder a child from cycling at higher intensities. Development of outcome measures like the smoothness and rhythm metrics is the first step toward quantitative assessment of motor control.

Both metrics are computationally inexpensive, clinically intuitive, and can be used to assess abrupt, jerky movements. More importantly, these metrics give us a snapshot of the cycling "quality" (e.g., irregular, halted, abrupt motion) over metrics that only measure cycling "quantity" (e.g., duration of cycling, cycling speed etc.). Thus, a child cycling with a smoother, more rhythmic motion after undergoing a rehabilitation program may demonstrate improved motor control rather than a child who may be cycling faster or for longer duration albeit with compensatory, maladaptive motions (e.g., backpedaling, arrested motion). If metrics to quantify the quality of motion are unavailable, then these compensatory motions may go unchecked and be reinforced over the training duration. The metrics in this study may help in identifying and targeting these deficits. For example, poor smoothness scores during cycling may indicate a need to address muscle spasticity and co-contraction in order to improve their cycling motion while poor rhythmicity may indicate a need to use metronomes or auditory cues at portions of the cycling revolution to ensure regular, rhythmic motion. Thus, these metrics may aid in designing rehabilitation programs to meet physical activity needs of not just children with cerebral palsy but other neurodevelopmental disorders as well. Additionally, the smoothness and rhythm thresholds derived from a decision tree analysis, potentially supported by a larger study, might serve as post rehabilitation targets for a cycling program for children with CP.

Due to impairments such as altered muscle activations patterns, agonist–antagonist co-contraction, and abnormal timing of activation during cycling, children with CP demonstrate an irregular, halted cycling pattern (19–21). Thus, they may be less likely to generate smooth and symmetric motion required to attain a high cycling intensities needed to attain cardio-respiratory benefits. The World Health Organization's International Classification of Function, Health, and Disability (ICF) model stresses the importance of incorporating a child's social and environmental needs into rehabilitation programs. Hence, it is critical to implement rehabilitation programs that incorporate functional activities that a child is personally motivated to perform and that improve their participation in family and social activities. Cycling provides a great way of addressing body structure and function components of ICF as well as encouraging participation in an activity that can be performed outside of the PT clinic using an adapted cycle with family and friends. The smoothness and rhythm metrics in this study provide an avenue to clinicians to quantitatively assess an "activity" rather than the traditional outcome measures that may be subjective or may evaluate a single plane movement. Improved ability to cycle smoothly and rhythmically may encourage participation of children with CP with their typically developing peers, siblings and friends in a socially enjoyable physical activity. Children with CP are more likely to participate in a physical activity if it lets them "fit in" and may be discouraged if the motor tasks are too challenging or make their disability or asymmetries in motion stand out (9, 30). Additionally, parents perceive symmetrical movements during physical activity as critical (30). By enabling smoother, rhythmic and in turn symmetric cycling motion, children may be more motivated to participate in a physical activity with higher confidence and self-esteem. Not only will this help in addressing the social development of children with CP but they can engage in an enjoyable activity that is not viewed as "exercise."

Lastly, it is important to note the "chicken and egg" problem of higher physical activity and smoother motion i.e. children with irregular and asymmetric motion are less likely to participate in physical activities whereas children with better motor abilities may find it easier to engage in physical activities (30, 31). Conversely, children with higher physical activity levels show better motor performance and motor learning abilities (32, 33) and hence, may have smoother, more rhythmic movements. We attempt to take the first step toward addressing this problem by developing metrics to analyze and with further development, correct such maladaptive motor behavior during cycling.

Limitations

There are some limitations to consider when interpreting the results of this study. Firstly, the data for each group were collected as a part of two separate studies and this may have introduced potential between group differences. While there were no significant differences between the ages for the two groups, overall the participants in the CP group were slightly younger than those in the TD group. The small difference in age combined with developmental changes occurring during the early teens and the onset of puberty may contribute to potential inter-group differences. Also, children with CP were asked to achieve a target cadence of 60 rpm while children with TD were asked to pedal as fast as they could. While the smoothness metric is unaffected by inter-participant differences in cycling cadences, the differences in cycling rhythm may be magnified or reduced. At this point, we do not know definitively the implications of the different cadences on the cycling rhythmicity and acknowledge it as a potential factor to consider when interpreting our results.

Secondly, an important limitation to consider is that because the study only looked at the differences in children with and without CP, which one might expect are more obvious, we do not know yet if these metrics can detect extremely small, subtle changes in smoothness and rhythm. Children with CP being novice cyclers might show starker differences when compared to children with TD, which may have had some previous experience of cycling. While we gave the CP group more practice sessions than TD to account for potential previous cycling experiences in participants in the TD group, the novelty of the cycling task for children with CP may still contribute to the lack of smoothness and rhythm seen in this group.

While the sample size of our study was relatively small, these results show that our smoothness and rhythm measures hold promise as novel outcome measures deserving of further study to quantify motor control during cycling in children with CP. The decision tree models explored here show potential for being able to classify CP vs. TD based on smoothness or rhythm. However, with so few samples, the threshold for separation that is derived from a nonparametric split along an axis is inherently coarse and variable. To gain confidence in a fitted threshold from this process, a much larger study is needed where we would expect greater density of observations in the region where a best split would occur and therefore a finer, less variable fitted threshold for classification. Future work with larger sample sizes and stratified sampling for GMFCS levels will be needed to establish the sensitivity and discriminatory ability of these metrics on a sample with different cycling and functional abilities. Additionally, future studies that establish testing criteria such as minimum required number of cycling revolutions in a trial, the effect of different cycling cadences particularly on rhythm will be beneficial to standardize the testing process for clinical use.

In summary, this study identified two novel objective measures for quantifying cycling performance by assessing smoothness and rhythm of cycling. These measures may indicate neuromotor differences during cycling in children with CP compared to their TD peers. In particular, significantly less smoothness and rhythm of cycling in children with CP as compared to TD might indicate poor timing and irregularity of movement, altered coordination and motor control. These measures are offered as potential markers for tracking progression of motor control deficits and maybe used to evaluate effects of intervention during cycling training in children with CP.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Institutional Review Boards at Temple University and University of Delaware. The patients/participants provided their written informed consent to participate in this study. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

AS and AB: conception and design of the work. AB, TJ, and SL: data collection and critical revision. AS, AB, and BB: analysis of data and interpretation. AS: drafting the work. AS, AB, TJ, BB, and SL: final approval of the work. All authors agree to be accountable for the content of the work.

FUNDING

This study was supported by NIH (R01HD062588), Shriners Hospitals for Children (Grant #8530), and from a Clinical Research Grant from the Pediatric Section of the American Physical Therapy Association.

ACKNOWLEDGMENTS

We would like to thank all the participants and their parents who contributed to the study.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fresc.2021.690046/full#supplementary-material

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Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers
in Rehabilitation Sciences

REVIEW
published: 21 September 2021
doi: 10.3389/fresc.2021.722668

What Is Intensity and How Can It Benefit Exercise Intervention in People With Stroke? A Rapid Review

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Background: Stroke is one of the major causes of chronic physical disability in the United Kingdom, typically characterized by unilateral weakness and a loss of muscle power and movement coordination. When combined with pre-existing comorbidities such as cardiac disease and diabetes, it results in reductions in cardiovascular (CV) fitness, physical activity levels, functional capacity, and levels of independent living. High-intensity training protocols have shown promising improvements in fitness and function for people with stroke (PwS). However, it remains unclear how intensity is defined, measured, and prescribed in this population. Further, we do not know what the optimal outcome measures are to capture the benefits of intensive exercise.

Aim: To understand how intensity is defined and calibrated in the stroke exercise literature to date and how the benefits of high-intensity training in PwS are measured.

Methods: A rapid review of the literature was undertaken to provide an evidence synthesis that would provide more timely information for decision-making (compared with a standard systematic review). Electronic databases were searched (including Medline, PubMed, CINAHL, and Embase for studies from 2015 to 2020). These were screened by title and abstract for inclusion if they: (a) were specific to adult PwS; and (b) were high-intensity exercise interventions. Eligible studies were critically appraised using the Mixed Method Appraisal Tool (MMAT). The data extraction tool recorded the definition of intensity, methods used to measure and progress intensity within sessions, and the outcomes measure used to capture the effects of the exercise intervention.

Results: Seventeen studies were selected for review, 15 primary research studies and two literature reviews. Sixteen of the 17 studies were of high quality. Nine of the primary research studies used bodyweight-supported treadmills to achieve the high-intensity training threshold, four used static exercise bikes, and two used isometric arm strengthening. Five of the primary research studies had the aim of increasing walking speed, five aimed to increase CV fitness, three aimed to improve electroencephalogram (EEG) measured cortical evoked potentials and corticospinal excitability, and two investigated any changes in muscle strength. Although only one study gave a clear definition of intensity, all studies clearly defined the high-intensity protocol used, with

Edited by:
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Specialty section:
This article was submitted to
Disability, Rehabilitation, and Inclusion,
a section of the journal
Frontiers in Rehabilitation Sciences

Received: *09 June 2021*
Accepted: *17 August 2021*
Published: *21 September 2021*

Citation:
Church G, Smith C, Ali A and Sage K
(2021) What Is Intensity and How Can
It Benefit Exercise Intervention in
People With Stroke? A Rapid Review.
Front. Rehabilit. Sci. 2:722668.
doi: 10.3389/fresc.2021.722668

most (15 out of 17 studies) clearly describing threshold periods of high-intensity activity, followed by rest or active recovery periods (of varying times). All of the studies reviewed used outcomes specific to body structure and function (International Classification of Functioning, Disability, and Health (ICF) constructs), with fewer including outcomes relating to activity and only three outcomes relating to participation. The reported effect of high-intensity training on PwS was promising, however, the underlying impact on neurological, musculoskeletal, and CV systems was not clearly specified.

Conclusions: There is a clear lack of definition and understanding about intensity and how thresholds of intensity in this population are used as an intervention. There is also an inconsistency about the most appropriate methods to assess and provide a training protocol based on that assessment. It remains unclear if high-intensity training impacts the desired body system, given the diverse presentation of PwS, from a neuromuscular, CV, functional, and psychosocial perspective. Future work needs to establish a clearer understanding of intensity and the impact of exercise training on multiple body systems in PwS. Further understanding into the appropriate assessment tools to enable appropriate prescription of intensity in exercise intervention is required. Outcomes need to capture measures specific not only to the body system, but also level of function and desired goals of individuals.

Keywords: stroke, exercise prescription, intensity, outcomes, international classification of function

INTRODUCTION

Worldwide, 15 million people suffer strokes each year of which 5 million die and another 5 million are permanently disabled (1). Within the United Kingdom, a stroke occurs in individuals every 5 minutes, affecting over 100,000 people each year, who join a population of 1.2 million people with stroke (PwS) (2). Stroke remains the fourth highest cause of death in the United Kingdom after dementia, ischemic heart disease, and respiratory disease.

A stroke occurs when there is a sudden insult to the central neurological system because the blood supply to the brain is impeded. It can lead to a number of physical, cognitive, and psychological difficulties. Severe hemiplegia presenting as unilateral paralysis of the arm and leg is the most common physical symptom, which in 57.7% of cases affects the right side of the body (3), with the upper limb being more severely involved due to the high proportion of strokes involving the middle cerebral artery (4). One week after the stroke, hemiplegia is still present in 89.1% of PwS, while at 1 month, 72% of individuals continue to experience unilateral weakness or hemiparesis (5). After 6 months, the incidence of hemiparesis is observed in at least 65% of PwS (6, 7). This paresis results in an inability to generate muscle strength that leads to abnormal posture, abnormal stretch reflex, reduced power production, and impaired voluntary movement (6).

PwS commonly present with pre-existing comorbidities that are already likely to compromise their CV function and fitness. PwS and those experiencing myocardial infarction demonstrate similar characteristics in relation to age of onset and prevalence of hypertension, hypercholesterolemia, heart failure, diabetes, and peripheral vascular disease (8, 9). Similar patterns are found in individuals presenting with diabetes as a comorbidity, resulting in changes to insulin resistance and changes to blood cellular biochemistry including the role of glucose transporter 4 (GLUT4) in facilitating glucose uptake to maintain control of blood glucose levels (10). These risk factors result in changes to CV fitness at rest and during submaximal exercise. This includes a reduced or preserved cardiac ejection fraction with reduced cardiac output, a reduced stroke volume, increased difference in arterial-venous oxygen levels, increased total systemic vascular resistance, reduced skeletal muscle mitochondrial density, and reduced skeletal muscle oxidative capacity (10–12).

These primary impairments post-stroke combined with potential comorbidities result in a further reduction of functional capacity through effects on metabolic function, immune and hormonal profile, and bone mineral density (12). Furthermore, this gives rise to a moderate to strong correlation with functional performance and gait velocity (13), with some authors reporting how a pathological gait of hemiplegia may have double energy costs compared to those of a healthy subject (14).

Exercise and physical activity play an important role in preventing and managing health conditions such as coronary heart disease, type 2 diabetes, stroke, mental health problems, musculoskeletal conditions, and some cancers. It also has a positive effect on well-being and mood, providing a sense of achievement or relaxation and release from daily stress (15). Physical activity has been defined as any bodily movement produced by skeletal muscles that requires energy expenditure. This may be playing, working, active transportation, house chores, and recreational activities (16). Social changes over the last 40 years and the impact of disabling disease are among the biggest factors affecting physical activity levels (17). This has resulted in the need for supplementary exercise when physical activity levels are low, with the Department of Health suggesting that where 150 minutes of moderate exercise is not feasible, then 75 minutes of vigorous intensity activity, shorter durations of very vigorous intensity activity, or a combination of moderate, vigorous, and very vigorous intensity activity should be used instead (18).

Exercise has become a long-term rehabilitation strategy for PwS where a combination of strength training and aerobic training has been demonstrated to increase functional capacity in day-to-day living (19). The impact of exercise can be captured using several outcome measures (20). The WHO International Classification Functioning, Disability and Health (ICF) (21) is a dynamic multidimensional classification of health and health-related domains. It considers: (1) body functions and structure (aspects of anatomy and physiology); (2) activities (actions and tasks undertaken by an individual); (3) participation (involvement in real-life situations); and (4) the environment and personal factors that may influence an individual. Within the healthy population, changes as a result of exercise intervention are usually captured by looking for changes to body structure and function, i.e., changes to ventilatory threshold and cardiac functioning. More recently, exploring the use of the WHO ICF (21) outcomes in clinical groups, has demonstrated a similar picture with a focus on outcomes of body structure and function and fewer outcomes focusing on activity and participation levels (22).

Elmahgoub et al. (23) set out how CV exercise occurs over three different intensity levels, low, moderate, and vigorous, which are measured by the Metabolic Equivalent of Task (MET). The effects of exercise at each intensity level result in a different training effect, with changes to VO_2, blood pressure (BP), blood lipid profiles, body mass index (BMI), blood glucose, mood, and quality of life (24).

In clinical practice, the percentage of maximal heart rate (MHR) or ventilatory threshold (VO_2) is commonly used to measure the intensity of CV training. Studies of healthy individuals have commonly used an intensity range from 70 to 95% of a MHR, ventilatory threshold, strength, or lactate threshold to achieve fitness changes. It remains unclear if using a lower range of 30–85% of MHR as an intensity guide yields proportionally lower changes to an individual (24, 25).

Higher intensity training has gained much popularity in the last 10 years due to the short-term benefits to walking speed, CV fitness, muscle strength, and changes to health biomarkers. Laursen et al. (26) identified changes in muscle enzyme activity in highly trained athletes, following high-intensity training. Despite no change in oxidative or glycolytic enzyme activity, there were significant improvements in endurance performance ($p < 0.05$). They also identified how increases in skeletal muscle buffering capacity may be one of the mechanisms responsible for an improvement in endurance performance. Changes in plasma volume, stroke volume, as well as myoglobin, capillary density, and muscle fiber characteristics have yet to be investigated in higher intensity training.

Mangine et al. (27) and Schoenfeld et al. (28) explored physiological changes to muscle physiology and structure with high-intensity strength training. Strength-focused training typically does not use MHR or VO_2 as a guide for intensity. Both strength-focused training and CV training rely on using a percentage of maximal power of an individual or strength production as a guide and working at a specific threshold of maximal intensity. Mangine et al. (27) and Schoenfeld et al. (28) identified changes to cross-sectional area, fiber type and size, pennation angles, and collagen content when comparing higher intensity with lower intensity training. They concluded that these changes are the most likely mechanism for improvement to fitness when compared with lower intensity training, despite both forms of training giving similar changes to metabolic functioning.

Optimal neuroplastic changes require a combination of skill, aerobic, and strength-based training to influence changes at cortical, subcortical, spinal, and peripheral levels of the nervous system (29). During neurological training, increasing the intensity of interventions appears to be one of the most beneficial components to improving functional performance (30). However, the definition of intensity, the aims of delivery, and the measurement of intensity in neurological or skills training is poorly understood and poorly standardized, when compared with CV and strength training (31–35).

It remains unclear if the underlying anatomical and physiological changes occurring during exercise as part of rehabilitation intervention at higher intensity positively affect all components of the WHO ICF framework. It also remains unclear if changes to outcomes to body structure and function (impairment), activities (limitations), or participation (restrictions) are affected, and if so, how is this captured in PwS (36).

Rapid reviews were introduced (37) to overcome a key barrier to the use of research evidence in decision-making (namely the delay in practitioners accessing and using research syntheses). In order to make the review rapid and timely, it restricts itself to studies that had been published recently (the last 5 years for example), excludes non-peer-reviewed work and unpublished/grey literature as well as avoiding non-English texts. A rapid review typically uses one reviewer only and has an optional quality assessment step (37, 38).

To date, there have been two systematic reviews covering high-intensity exercise for PwS (39, 40), which looked solely at what exercises were used in high-intensity training. Neither of these gave a clear definition for intensity and did not explore the aims of the intervention. This review therefore intends to fill these gaps in the exploration of intensity. The aims of this review are set out below:

1. Explore how intensity is defined within the exercise interventions for PwS.
2. Document the aims of the exercise interventions for PwS (e.g., cardiovascular function, muscle strength etc.).
3. Identify the methods/tools used to measure intensity during the exercise interventions.

TABLE 1 | Eligibility criteria for inclusion and exclusion and their justification.

Code	Inclusion	Exclusion	Justification for rapid review Haby et al. (37) and Dobbins (38)
1	Peer-reviewed literature from 2015 onwards	Peer-reviewed papers prior to 2015 Unpublished/draft publications Grey literature	Ensure up to date literature is reviewed and excludes literature that has not undergone peer review.
2	Intensity-specific exercise intervention	Intensity not part of the intervention Non-exercise specific	Intensity only literature
3	Describes method used to deliver intensity	No description of the method used to deliver intensity	Intensity delivery methods must be identified.
4	Stroke specific clinical group	Non-stroke population (health/other clinical groups) Stroke data cannot be disaggregated from other clinical populations	Review is specific to PwS and therefore other clinical groups and non-clinical groups have been excluded.
5	Participants 18 and over	Participants under 18	Excludes participants under 18 where physiological response to exercise may differ.
6	Human studies	Not involving humans	Ensures findings are generalizable to human participants.
7	Articles written in English	Non-English articles	Avoids translation time and costs needed for foreign studies.

4. Document how studies prescribe intensity in the exercise interventions for PwS and how intensity is monitored during exercise.
5. Identify outcome measures used to capture change as a result of exercise training in stroke and whether these are mapped across the WHO ICF constructs.

METHODOLOGY

This study used guidance on the methodological process for a rapid review from Haby et al. (37) and Dobbins (38). The rapid reviews involve one reviewer and use strict eligibility criteria when selecting articles.

The sequential steps for this review are based on the Search, Appraisal, Synthesis and Analysis (SALSA) elements (41):

- Systematically search the literature and identify appropriate papers for the rapid review.
- Appraise the quality of papers using an appropriate quality assessment tool.
- Synthesize the content to identify themes and patterns.

The eligibility criteria followed those of Haby et al. (37) and Dobbins (38) and can be viewed in **Table 1**.

Four databases (Medline, Pubmed, CINAHL and Embase) were searched in November 2020. Searches were restricted from January 2015 to November 2020.

A building block approach (42) identified search terms for each concept. The concepts were: exercise (Concept A); stroke (Concept B), and intensity (Concept C). The search strategy comprised:

(a) Terms to describe stroke
(b) Terms to describe exercise
(c) Terms to describe intensity

These are shown in **Table 2**.

TABLE 2 | Example of search strategy including concepts, key words and MeSH terms.

Concept A	Concept B	Concept C
MeSH Subject heading	**MeSH Subject heading**	**MeSh Subject heading**
MeSH "Exercise+ or Activity+"	MeSh "Stroke+ or Cerebral Haemorrhage+" Stroke* or CVA or cerebrovascula* acciden*	MeSH "Intensity or intense"
OR	**OR**	**OR**
Keywords	**Keywords**	**Keywords**
Physica* activ* or physical exert* or exercis* therap*	post stroke or cerebrovascular or cerebral hemorrhage or cerebral vascula*	High-intensity or High-intensity interval training or HIIT or Moderate intensity interval training or MIIT
OR (specific terms for types of exercise)	**OR (result of or impact outcomes of stroke)**	**OR (Less commonly used)**
Exercise* or train* or strength* or strength* or isometric* or aerobic*. or endurance* or weigh* resist* or train or run*or job*. or walk*. or resistance* train* or Program*	TIA or transient isch* or infarct*or brain isch?emi* or aphasi*, Heminopia, Cognition?	
Search set A AND	**Search set B**	**AND Search set C**

MeSH, keyword, and specific term searches were completed. The Boolean operators AND and OR were to be used, alongside phrase, proximity, and truncation operators dependent on the database used. The search syntax was adapted for each information source and controlled vocabulary terms used where available.

Screening of papers on title and abstract was undertaken by the first author to identify those that met the inclusion and exclusion criteria. The first author then excluded papers by reading the full text. Ten of the papers excluded were sent to a second author (KS) for verification.

The extraction tool was developed and piloted by the first author (GC) on 10% of the papers. These were then checked and agreed by a second reviewer (KS).

The extracted data included basic information (authors, year of publication, type of paper, and location). In addition, more specific information to achieve the review outcomes included finding:

- Definitions of intensity
- Which body system the exercise was aimed at (e.g., CV system, muscular system, or neurological system)
- Measurement tools used in the assessment of intensity [maximal ventilation (VO_2 max)/gas exchanges testing, rate of perceived exertion (RPE), HR, repetition maximum, functional outcomes, and patient reported outcome measures (PROM)]
- How intensity is prescribed in PwS
- Outcome measures used to quantify the effect of exercise at various intensities on the body (resting HR and BP, blood lipid profiles, VO_2 max, 6-min walk test (6 MWT), shuttle run test, etc.)

Full-text articles identified as eligible during screening were then assessed for quality using the Mixed Methods Appraisal Tool (MMAT) (43). Papers were not excluded on the basis of the quality assessment. The quality assessment of studies provided an indicator of the robustness of the studies included in the review.

Narrative synthesis, with supporting tabular synthesis, drew together the information on:

- Homogeneity or heterogeneity in the terms used to define intensity within papers collected.
- Methods used in the testing and assessing fitness in PwS.
- Clarity of exercise-intensity prescription when used as an intervention and rehabilitation technique.

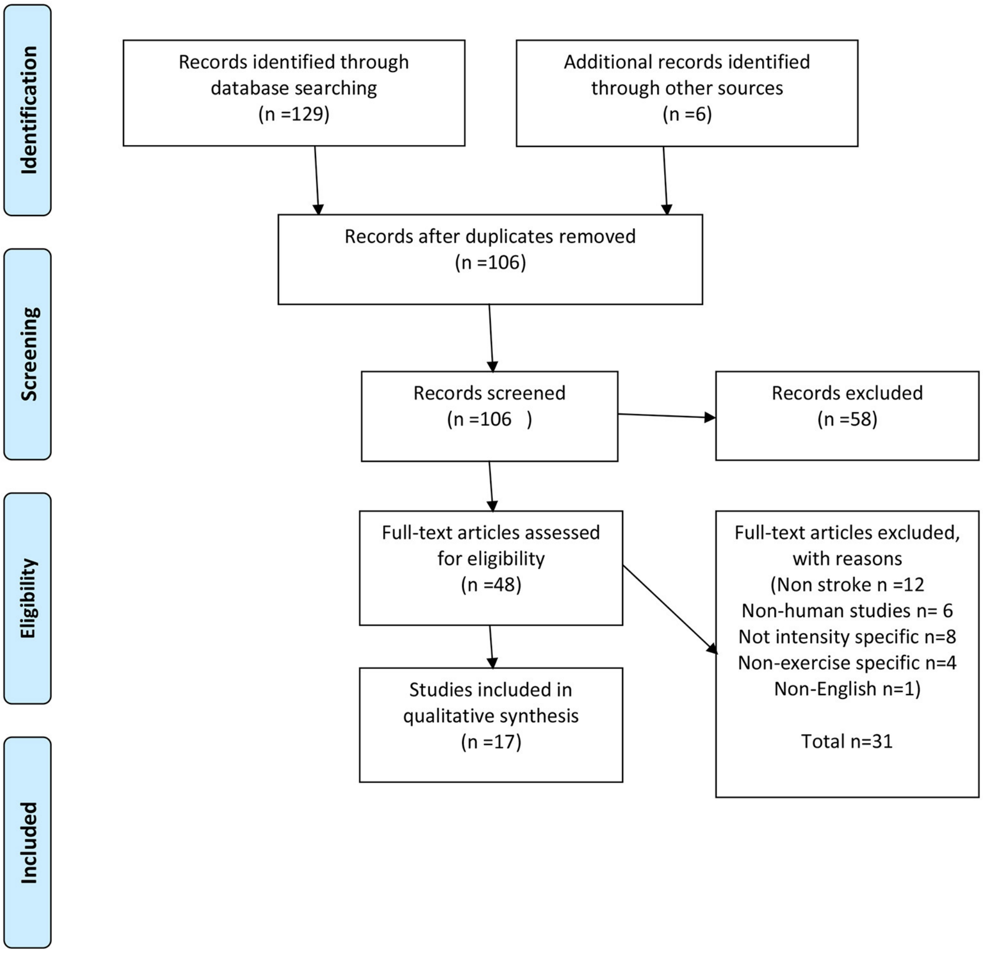

FIGURE 1 | Prisma flow diagram.

- How intensity impacts on changes to outcomes and what outcome measures within the constructs of the WHO ICF framework were commonly used.

RESULTS

Database searches found 129 records with an additional six from a reference list review. After duplicates were removed, 106 records were screened and 48 were screened by title and abstract for full-text eligibility assessment, leaving 17 articles for the purpose of this review. Of the articles excluded, 12 were non-stroke-related, 6 did not involve humans, 8 were not intensity-specific, 4 were non-exercise-specific, and 1 was only available in Chinese. A full breakdown of this process is included in **Figure 1** (PRISMA flow diagram).

The 17 articles were subjected to MMAT evaluation; eight were quantitative non-randomized controlled trials (RCTs), eight were quantitative RCTs, and two were qualitative reviews of the literature. Eleven papers came from the Unites States, two from Canada, and single records from Norway, Germany, Denmark, China, and Italy. Participant numbers ranged from 6 to 36 in the primary studies reviewed.

The MMAT quality assessment indicated that all the articles had clear research questions, appropriate and clear data collection methods to address the question and approach, interpretation of the data, and coherence during the analysis and synthesis. Only one of the RCTs (44) failed to identify if assessors were blinded for the process.

Table 3 summarizes the findings from the included studies.

Homogeneity and Heterogeneity

The definition of intensity was only identified in one paper as "the work rate, effort level, or metabolic demand of aerobic activity quantified by heart rate, rate of oxygen consumption, rating of perceived exertion and/or walking speed" (45).

Collectively, the reviewed studies identified working at or above 80–90% of MHR, VO_2, or one repetition maximum of an individual classifies as a high-intensity intervention, where moderate intensity was aimed toward 40–60% of these physiological outcomes. This does vary between studies and does not always consider rest intervals or ratio as used in Boyne et al. (44). The studies found by this review all focused on higher intensity exercise interventions. This may reflect the current trend in researching potential health benefits of higher intensity exercise for clinical and non-clinical groups when compared with lower intensity training (27, 28).

All of the primary research studies identified clear objectives for how they used high-intensity training protocols. Munari et al. (45) was the only study that discussed and defined intensity and the impact of intensity on participants. Neither review study (39, 40) provided definitions of intensity but shared similar findings to the primary research studies in relation to the intensity levels used in high vs. moderate exercise interventions.

Desired Training Effects

Multiple desired training effects were sought in primary studies and reviewed in both of the systematic review studies. The most common intended training effect was improved walking speed using bodyweight supported treadmill training, used in 14 of the 17 (82%) studies (39, 40, 44–49). Improved CV fitness was used in 13 of the studies (76%) (39, 40, 44, 50–53). Changes to brain activity measured by an electroencephalogram (EEG) was reported in 5 (29%) of the 17 studies (47, 50, 54–56).

Methods Used in Testing and Assessing Fitness in PwS

Intensity assessment was achieved using a graded exercise test (GXT) in 11 of the 15 (73%) primary research studies to obtain a predicted maximal oxygen consumption (VO_2) and MHR measure (44, 45, 54, 57) and age-predicted MHR calculation (45, 48, 49, 54, 56). Two used maximal strength testing (55, 58), one used a home-based walking test and RPE to establish exercise effort (52), and one used age-predicted values for MHR and VO_2 (56). Both of the review studies (39, 40) shared consistent findings with the primary studies in this review for the methods used to assess intensity level for interventions.

Intensity Prescription and Within-Session Monitoring

All primary research studies used high-intensity exercise prescription with the effects captured over a maximum of 3 months (45). The two systematic review studies were consistent with this finding. Intensity progression within studies was prescribed most commonly using walking speeds (10 of 17 studies) (39, 44, 46–48, 50, 51, 56, 58). The RPE using the BORG 6-20 scale was used as a method of prescribing exercise intensity in 6 (29%) of the 17 studies (45, 47–49, 54, 56), while only one study used mixed methods for intensity prescription combining walking speed, percentage VO_2, and recovery interval timings (39).

The monitoring of within-session intensity exercise using MHR was the most common method employed (10 studies) (44, 45, 47–49, 51, 54, 56, 57). The use of RPE and BORG 6-20 scales were also commonplace (nine studies) (44, 45, 47–49, 52–54, 56). Neither of the systematic review papers in this review reflected on the within-session monitoring methods during exercise interventions.

Outcome Reporting

All of the primary research studies used outcome measures relating to body structure and function (as defined in the WHO ICF checklist) including VO_2, HR, BP, blood lipids, blood biomarkers, interleukins, corticospinal excitability, and electromyography. Of these, Högg et al. (58) and Krawcyk et al. (52) used outcomes identified by Salter et al. (36) as reliable, valid, and responsive to change in PwS. **Table 4** shows the outcome measures and how they line up with the WHO ICF constructs.

Five studies (51, 54–57) failed to use outcome measures related to activity, 12 (70%) used the 6 MWT, 5 (29%) used 10-meter walk test (10 MWT), and 4 (12%) used walking speeds obtained from the treadmill.

Four studies used outcome measures relating to participation (45, 48, 52, 58), of which two measures were recommended by Satler et al. (36), the Stroke Impact Scale and Short Form—36.

TABLE 3 | Summary results of included papers: type of study, training effect, methods for increasing intensity.

First name author	Paper title	Journal	Date	Location	Type of study*	Intended training effect	Method of assessing intensity	Method of monitoring intensity within session	Methods to progress intensity
Aaron et al.	Feasibility of single session high-intensity training utilising speed and active recovery to push beyond standard practice.	Topics in Stroke Research	2018	USA	Quant Non-RCT	Walking speed	Walking speed on treadmill	Walking speed and quality	Incremental walking speed
Abraha et al.	A bout of high-intensity interval training lengthened nerve conduction latency to the non-exercised limb in chronic stroke.	Frontiers in Physiology	2018	Canada	RCT	Cardiovascular (CV) fitness, strength, upper limb function and cognitive timing	Maximum (VO_2) max testing	%VO_2 max	Increasing %VO_2 and walking gradient
Boyne et al.	Within-session responses to high-intensity interval training in chronic stroke.	Clinical Sciences	2015	USA	Quant Non-RCT	CV fitness and walking speed	(GXT) for MHR for MHR and VO_2	Walking speed and % maximal effort from % of GXT	Increasing walking speed and gradient
Carl et al.	Preliminary safety analysis of High-intensity interval training (HIIT) in persons with chronic stroke.	Applied physiology, Nutrition and Metabolism	2016	USA	Quant non-RCT	Safety	GXT for MHR and VO_2	ECG	Reduced recovery times
Crozier et al.	High-intensity interval training after stroke: an opportunity to promote functional recovery, cardiovascular health and neuroplasticity.	Neurorehabilitation and Neural Repair	2018	Canada	Qualitative review	CV fitness and walking speed	VO_2 max testing and walking speed	N/R	Variation of increasing %VO_2 max, walking speed, recovery time
Gjellesvik et al.	Effects of high-intensity interval training after stroke (The HIIT stroke study)	Archives of physical medicine and rehabilitation	2020	Norway	RCT	CV fitness	VO_2 max testing	% MHR	Increasing walking speed and gradient
Högg et al.	High-intensity arm resistance training does not lead to better outcomes that low intensity resistance training in patients after sub-acute stroke	Journal of rehabilitation medicine	2020	Germany	RCT	Upper limb strength and function	1 Repetition Maximal (RM) functional strength testing for upper limb	Range of motion and repetitions completed	Increasing range of motion and repetition until achieving 15
Krawcyk et al.	Effect of home-based high-intensity interval training in patients with lacunar stroke.	Frontiers in Neurology	2019	Denmark	RCT	CV fitness, meatal health and well-being, Body mass index and activity levels	Talk testing	RPE (BORG 6-20)	RPE (BORG 6-20)
Leddy et al.	Alterations in aerobic exercise performance and gait economy following high-intensity dynamic stepping training in persons with sub-acute stroke.	Journal neurological physical therapy	2016	USA	RCT	CV fitness and walking speed.	GXT testing for MHR and VO_2	%MHR, gait quality, RPE (BORG 6-20)	%MHR and RPE (BORG 6-20)

(Continued)

TABLE 3 | Continued

First name author	Paper title	Journal	Date	Location	Type of study*	Intended training effect	Method of assessing intensity	Method of monitoring intensity within session	Methods to progress intensity
Li et al.	A short bout of high-intensity exercise alters ipsilesional motor cortical excitability post stroke.	Topics in Stroke Rehabilitation	2019	USA	Quant non-RCT	Brain activity	Age predicted calculated MHR	%MHR and RPE (BORG 6-20)	Progressive walking speed to achieve target %MHR
Luo et al.	Effects of high-intensity exercise on cardiovascular fitness in stroke survivors.	Annals of Physical and rehabilitation medicine	2020	China	Qualitative review	CV fitness and walking speed.	Not discussed	N/R	N/R
Madhavan et al.	Effects of single session of high-intensity interval treadmill training on cortical excitability following stroke.	Journal of neural plasticity	2016	USA	Quant non-RCT	Brain activity and walking speed.	10-meter times walk	%MHR, RPE (BORG 6-20), blood pressure	10% increase in walking speed if able to tolerate previous session
Madhaven et al.	Effects of High-intensity speed-based treadmill training on ambulatory function in people with chronic stroke: A preliminary study with long term follow up.	Scientific Reports	2018	USA	Quant Non-RCT	Walking speed	Age predicted calculated MHR and 10-meter walk test	%MHR, RPE (BORG 6-20) and gait quality	Progressive increase from 50% walking speed until exceeding 80% MHR or gait disturbance
Mahtani et al.	Altered sagittal and frontal plane kinematics following high-intensity stepping training versus conventional interventions in sub-acute stroke.	Physical Therapy	2017	USA	RCT	Walking quality of movement	Age predicted calculated MHR and RPE (BORG 6-20)	RPE (BORG 6-20), %MHR and BP	%MHR and RPE (BORG 6-20)
Munari et al.	High-intensity treadmill training improves gait ability, VO_2 and cost of walking in stroke survivors: preliminary results of a pilot RCT.	European Journal of Physical and Rehabilitation Medicine	2018	Italy	RCT	Walking quality of movement	Age predicted calculated MHR and Borg 6-20 PRE	%MHR and RPE (BORG 6-20)	%MHR and RPE (BORG 6-20)
Nepveu et al.	A single bout of High-intensity Interval training improved motor skill retention in individuals with stroke.	Neurorehabilitation and Neural Repair	2017	USA	Quant non-RCT	Brain activity.	GXT for MHR and VO_2 with age predicted MHR	%MHR and RPE (BORG 6-20)	Participants working at 100% maximal walking speed- no progressions made
Urbin et al.	High-intensity unilateral resistance training of a non-paretic muscle group increases active range of motion in severely paretic upper extremity muscle group after stroke.	Frontiers in Neurology	2015	USA	Quant non RCT	Brain activity, strength and range of motion	1RM for isometric resistance strength	ROM and observed fatigued onset	Increasing range of motion and %1RM

**Using MMAT definition.*

N/R, Not reported; quant, quantitative; RCT, Randomized Control Trail; GXT, Graded Exercise Testing; RPE, Rate of Perceived Exertion; MHR, Maximal Heart Rate; VO_2, Ventilatory oxygen threshold; RM, Repetition Maximum; BP, Blood pressure; CV, cardiovascular.

TABLE 4 | Outcomes measure linked to WHO international classification of functioning, disability, and health constructs.

First name author	Outcomes to measures used and relation to WHO ICF checklist		
	Body structure and function	**Activity**	**Participation**
Aaron et al.		Walking speed on treadmill	
Abraha et al.	Maximum ventilatory threshold (VO$_2$), Heart Rate (HR), Motor Evoked Potentials (MEP), Corticospinal Excitability (CSE), grip strength	***Box and block test***	
Boyne et al.	Exercise tolerance (completion of the 20 min session), VO$_2$, HR	Walking speed on treadmill	
Carl et al.	Electrocardiogram (ECG)		
Crozier et al.	VO$_2$, HR, MEP, Blood Pressure (BP)	***6-min walk test***, 10 MTW, ***Berg balance test***	
Gjellesvik et al.	VO$_2$, BP, Blood profiles including High Density Lipoproteins (HDL), triglycerides, Glycated Haemoglobin (HbA1c), C-peptides		
Högg et al.	Grip strength, Motricity index**,** ***Fugl-Meyer assessment, modified ashworth scale***	Goal Attainment Scale (GAS)- specific to activity of an individual, ***Box and block test***	GAS- specific to participation of an individual
Krawcyk et al.	Endothelial function (plethysmography), hyperaemia index, HR and augmentation index, BP, multiple biomarkers (Pro-adrenomedullin, Pro-atrial natriuretic peptide, inter leukin 6, Tumour necrosis factor, ICAM-1 protein, VCAM-1 Biomarker, vascular endothelial growth factor. BMI. Multidimensional Fatigue Inventory (MFI-20 questionnaire), Major Depression Inventory (MDI), World Health Organisation Five well-being (WHO-5), Chronic stress Ull-meter, ***Montreal Cognitive Assessment***, Metabolic Equivalent of Task (MET) calculations from activity and HR measures,	Daily steps using accelerometer	Physical activity levels *via* Physical Activity Scale V2
Leggy et al.	VO$_2$, MHR, oxygen cost walking from VO$_2$	***6 MWT***	
Li et al.	EMG, TMS		
Luo et al.	VO$_2$ and VO$_2$ peak, pain VAS, injury rates	***6 MWT***, 10 MWT, Falls frequency,	
Madhavan et al.	Electromyography (EMG), Transcranial Magnetic Stimulation (TMS)	walking speed, 10 m walk	
Madhaven et al.	HR. BP	10-meter timed walk, **6 MWT**	***Stroke Impact Scale (SIS)***
Mahtani et al.	HR, BP, Range of motion	Stepping symmetry, gait speed,	
Munari et al.	VO$_2$, oxygen cost of walking, HP, BP,	10 MWT, ***6 MWT***, ***TUAG***	***SF-36 and SIS***
Nepveu et al.	TMS for CSE and Intra Cortical excitability, MVC,		
Urbin et al.	EMG, TMS, range of motion,		

Outcomes in bold and underlined represent those specific to stroke from the ICF WHO Evidence Based Review of Stroke Rehabilitation (EBRSR) as identified and reviewed in (36). Bold represent outcomes from the ICF used in PwS.

Six studies (45, 48–50, 53, 58) used outcome measures identified in Salter et al. (36), the most common of these being the 6 MWT. Only one of the systematic review studies (39) explored outcomes specific to PwS in exercise interventions. They also found that the 6 MWT was the most commonly used outcome and suggested this was due to the practicality and functional relevance for PwS.

Munair et al. (45) was the only study to explore the safety surrounding the use of high-intensity training intervention in stroke. Safety appeared to be supported in all studies as there was no mention of adverse events or dropouts of study participants. Neither of the systematic review studies explored safety issues.

DISCUSSIONS

This review has appraised a range of high-intensity interventions for PwS, which aim to increase CV fitness, improve muscle strength, increase functional capacity, or to increase brain activity.

Homogeneity or Heterogeneity in Defining Intensity

All of the studies identified clear objectives and protocols of how they used high-intensity training. Munari et al. (45) was the only study that discussed and defined intensity and the role of intensity in interventions as the work rate, effort level, or metabolic demand of aerobic activity quantified by HR, rate of oxygen consumption, rating of perceived exertion, and/or walking speed. Despite various definitions of intensity in exercise interventions in non-clinical groups, there was no clear definition of intensity during exercise intervention in PwS. It remains unclear if defining intensity shares similarities or differences if the exercise intervention is aimed at a specific body system such as CV system compared with interventions aimed at improvements in a functional task such as walking speed.

The Body System the Intervention Was Aimed At

Primary studies in this review and the two systematic review studies all had a key aim for exercise. This varied from improving CV fitness through changes to MHR or VO_2 (44, 50), improving functional capacity through changes to walking speed (47, 48), and changes to brain activity *via* increased cortical firing rates (54). None of the studies sought to establish whether changes to a body system such as CV fitness actually resulted in improvement to function, or whether training a functional task such as walking would have differential impacts on the neurological, musculoskeletal, or CV systems.

Findings relating to intervention aims to support the Specific Adaption to Imposed Demands (SAID) principles identified in Sale et al. (59). These principles identify how the human body will adapt to any demand whether the stressor is biomechanical such as muscular, CV, or neurological. This can be observed in all of the primary studies that used treadmill training (44, 46–49, 53), where there is an identified improvement to walking function, but not necessarily changes in muscle strength, CV fitness, or motor potential.

Methods Used in the Testing and Assessing Fitness in PwS

The assessment of intensity in studies ranged from the gold standard in healthy populations VO_2 max testing (50) to graded exercise testing (44), RPE (53), and the talk test (52). Using graded exercise testing, percentage MHR from age-predicted value or obtained from GXT and RPE as assessment procedures are more practical and transferable to clinical practice. The studies were not consistent with their choice or reasoning for the assessment tool used. It was also unclear if using an assessment intervention such as percentage MHR or RPE using the BORG 6-20 scale showed any correlation with ability to achieve a percentage MHR in activities such as treadmill walking. No account was taken of other limiting factors e.g., lower limb strength rather than the CV demand of walking.

There was no clear consensus about an appropriate method for capturing baseline fitness of an individual. This was demonstrated by Munair et al. (45) who discussed intensity and its role in exercise prescription and how this needs to be specifically aimed at the appropriate body structure, functional task, or energy system the exercise intervention is being aimed at. They also discussed how other systems such as muscular strength or power may limit an individual reaching the desired level of intensity from a CV perspective.

Protocols for Exercise-Intensity Prescription

A variety of methods were used to deliver an intensive intervention. These included achieving target percentage MHR or VO_2 (53), percentage maximal walking speed (45), percentage of one repetition maximum (58), or adjusting recovery periods to a ratio or working intervals (44). There was some consistency in papers reviewed that working at or above 80–90% of MHR, VO_2, or one repetition maximum of an individual classifies as a high-intensity intervention. It may be that a variety or combination of methods could be used to ensure sessions are high in intensity (39) and would be similar to periodization programs described by Lorenz et al. (60), where intensity of sessions is progressed in an undulating linear fashion allowing deloading or recuperation days. However, there was a lack of standardization for developing high-intensity training protocols. Different methods may create different outcomes or more specifically, certain methods used to create high-intensity sessions could be tailored depending on individuals pre-assessment fitness and ability findings. Eng et al. (13) and Flansbjer et al. (61) highlighted the importance of muscle strength in the performance of functional tasks and specifically correlations between lower extremity muscle strength and gait performance and how this relates to an increased perceived ease of participation during functional tasks.

No studies considered the long-term (more than 3 months) effects of short duration high-intensity training compared with the long-term effects of lower intensity longer duration training or higher volume training. In healthy adults, high-intensity strength-based training interventions and high-intensity anaerobic interventions demonstrate changes to body structure and function sharing similarities to aerobic training in relation to molecular signaling pathways (20). They also cause changes to muscle structure relevant to the stroke population such as improved pennation angle and sarcomere development. Future studies on exercise in stroke would benefit from investigating long term follow-up and combined interventions at various intensities to optimize protein synthesis and muscle architecture, potentially further enabling functional capacity in PwS.

Within-Session Monitoring of Intensity

Within-session monitoring of individuals appeared appropriate to the intervention. Studies using a CV intervention such as treadmill training or cycling would typically use CV markers such as HR or VO_2 calculations as a guide. RPE was one of the most commonly used methods with studies selecting the BORG 6-20 scale. Of the 17 included papers, only Krawcyk et al. (52) discussed the methods used to calibrate RPE (using the BORG 6-20 scale) and how this correlates to HR. None of the studies discussed the possible issues surrounding the inter-rater reliability of using a subjective perceptual scale, or if the perceived effort score is related to dyspnea or muscle fatigue.

The majority of the studies used a CV intervention to create changes to the CV system thereby increasing the functional capacity to walk further or more quickly. Some studies have used CV high-intensity training to investigate the benefits of brain activity by increasing cerebral blood flow (55, 56). Studies typically use electroencephalography (EEG) and transcranial magnetic stimulation (TMS) to evaluate the changes to brain activity. Both failed to evaluate if the intensity needed to create this change from a CV intervention was sufficient or appropriate for increasing brain activity as it was for increasing CV fitness.

How Intensity Impacts of Changes in Exercise Intensity and How these Relate to Outcomes Within the Constructs of the WHO ICF Framework

All of the studies considered the use of outcome measures related to the body structure and function. This has been defined in the ICF checklist as the anatomical parts of the body and the physiological functions of body systems. These measures are keys for identifying the changes to CV fitness, muscle strength changes, and cortical excitability. While these changes may be of importance to elite athletes who are looking for the smallest of changes in competition, changes to functional activity and participation are thought to be more important to PwS (62). Outcome measures relating to activity and participation have been defined in the ICF checklist as the execution of a task or action by an individual i.e., stair climbing and the involvement of those tasks in real life situation i.e., climbing the stairs three times a day to use the toilet. This challenges the applicability of the studies for rehabilitation as their outcomes were not goal centered for PwS. None of the studies reviewed considered using this approach and despite the best efforts to ensure individuals are exercising at a specific intensity, it may be challenged that individual motivation may vary and could impact on their participation effort.

Stroke specific, validated outcome measures were used in eight of these studies. Salter et al. (36) assessed reliability, validity, and responsiveness of outcome measures in stroke. This review found functional testing such as the 6 MWT and Timed Up and Go (TUG) test were the most commonly used. Using outcome measures such as these, which may be more reliable, valid, and responsive and maybe more meaningful to the participant, might capture greater improvement from the intervention. None of the studies reported whether there was any education provided to the participants to help them understand the benefits of higher intensity training protocols.

All of the studies demonstrated a beneficial effect for high-intensity training on body systems, activities, and outcome measures. The limited use of functional outcome measures may be a factor in compliance and motivation in interventions. Högg et al. (58) used goal attainment scaling (GAS) of individuals to allow the individuals to select appropriate goals for fitness/activity/movement improvements. Sixty percent of the higher intensity and 55% of the moderate intensity group achieved their participation-specific GAS outcomes. All groups increased in grip strength and most experienced no changes to spasticity.

While there was an identified need to use a harness in walking intervention as a safety precaution, there was minimal discussion about safety and the need to tailor exercise to meet the specific needs of each individual with stroke, and there was no record of how the needs of PwS were addressed when there were issues.

Comparison to Previous Literature

Nichols et al. (63) report that exercise intensity in cardiac rehabilitation programs can be suboptimal. This may limit potential intervention benefits on neuroplasticity, strength, and CV fitness in programs treating PwS. Neurological training specifically lacks an appropriate methodology to measure intensity during skill training (19) and as a consequence, potential neuroplastic gains made through skill or sensory-motor training, strength training, and CV training in individuals may not be optimal.

Due to the high-intensity nature of the studies used in this review and lack of short term follow-up, it remains unclear if increasing the intensity of exercise provides any significant long-term physiological, physical, or psychological benefits to PwS over and above high volume-low-intensity training. None of the studies in this review identified how individuals need the sufficient support systems such as the neurological function and muscle power to participate in varied CV-based interventions, something that is taken for granted in non-clinical groups or clinical groups who do not have significant physical impairments. Furthermore, rigorous assessments of all body systems would allow for an appropriate selection of assessment tools to establish tailored intensity levels or thresholds for the desired body system. Methods of monitoring the intensity of the session specific to the intervention and using specific and sensitive outcome measures to detect changes at all levels of the ICF is key. More specifically, the need to identify benefits tailored to an individual.

This review has identified that changes to mood and quality of life can be related to the physiological changes brought about by the exercise component. None of the reviews acknowledged the potential social benefits of exercise participation, which might bring about improvements to mood and quality of life (57).

This review also did not identify how additional strength gains may not be associated with further improvement in an activity (5). Strengthening beyond the functional needs of an individual may be of value for establishing a functional reserve rather than further improvement in current performance at a functional activity. This may also be the case with neurological/skill training and CV-based training (64).

CONCLUSIONS

This review has explored the use of intensity in exercise training intervention for PwS, and how this varies depending on the desired effect on the body system or task-specific activity. The tools most employed to gauge exercise intensity and that can be translated to clinical practice for monitoring intensity are MHR and BORG RPE. Despite this, there is a lack of consensus about how to define exercise intensity across CV, muscular strength, neurological, and functional skill training, and how this is applied in a meaningful way to PwS to optimize the benefits. A clear understanding of intensity is essential to focus the desired training effect required in exercise interventions and improve the prescription of intensity by therapists and exercise prescribers. More focus on the desired effect would allow the appropriate intensity training methods to be selected and consideration given to whether longer duration, moderate intensity training should be combined with higher intensity training for optimal benefits.

A thorough understanding of the needs of the PwS, specifically the multidimensional issues they present with, is required in order to tailor the intensity, type of exercise, and methods of training. Although not covered in this review, the need for education related to the intervention needs to be considered when selecting outcome measures. This includes the exercise desired effects at a physiological level and how this can be used to improve meaningful outcomes such as skill reacquisition. This can then be used to allow PwS to see how these benefits can impact on the achievement of everyday tasks and furthermore into the reintegration into social participation.

Finally, we need a better understanding of the timescales required for exercise interventions to make the desired changes in PwS. It is unlikely that single bout interventions are able to provide a meaningful snapshot of the actual benefits of varied exercise-intensity interventions. Additionally, if there are superior health-related benefits with higher intensity training, further consideration is need about the effect on long-term adherence compared to lower intensity exercise interventions in PwS.

AUTHOR CONTRIBUTIONS

GC, KS, AA, and CS: conceived and designed the study. KS, CS, and AA: critical review of paper. GC: writing of paper, analysis, interpretation of findings, and conducting literature searches. GC and KS: piloting data collection and quality measure. GC, CS, and KS: inclusion and exclusion process. All authors contributed to the article and approved the submitted version.

FUNDING

GC was funded by Health Education England (HEE)/National Institute for Health Research (NIHR) on a Pre Doctoral Research Fellow (NIHR300426).

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fresc.2021.722668/full#supplementary-material

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Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers
in Rehabilitation Sciences

HYPOTHESIS AND THEORY
published: 01 October 2021
doi: 10.3389/fresc.2021.710618

Flipping the ICF: Exploring the Interplay of Theory and the Lived Experience to Reconsider Physical Activity in Community-Dwelling People With Multiple Sclerosis

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People with multiple sclerosis (MS) report lower physical activity levels and are at a risk of becoming sedentary. As such, they are at an increased risk of developing secondary health conditions associated with inactivity. This is of major public health concern. Attempts to improve the physical activity levels in people with MS remain a challenge for health professionals. One key reason might be the lack of understanding about the meanings people with MS ascribe to exercise and physical activity. This paper draws on the key findings of a three-phased interconnected mixed methods sequential explanatory study to examine the meanings of exercise and physical activity from the perspectives of people with MS and health professionals. Phase 1 used a four-round Delphi questionnaire to scope and determine the consensus of priorities for exercise and physical activity and the reasons why people with MS ($N = 101$) engaged in these activities. Phase 2 used face-to-face semistructured interviews of people with MS ($N = 16$) to explore the meanings ascribed to exercise and physical activity. Phase 3 explored the perceptions of physiotherapists ($N = 14$) about exercise and physical activity using three focus groups. Using the International Classification of Functioning, Disability, and Health as a theoretical framework to underpin this study, the authors discuss the key factors, for example, emphasis on the contextual factors, that drive decision making around exercise and physical activity participation in people with MS and explore the clinical implications to health professionals.

Keywords: exercise, physical activity, priorities, ICF (international classification of functioning disability and health), physiotherapy, multiple sclerosis

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Specialty section:
This article was submitted to Disability, Rehabilitation, and Inclusion, a section of the journal Frontiers in Rehabilitation Sciences

Received: *16 May 2021*
Accepted: *16 August 2021*
Published: *01 October 2021*

Citation:
Stennett AM, De Souza LH and Norris M (2021) Flipping the ICF: Exploring the Interplay of Theory and the Lived Experience to Reconsider Physical Activity in Community-Dwelling People With Multiple Sclerosis. Front. Rehabilit. Sci. 2:710618. doi: 10.3389/fresc.2021.710618

INTRODUCTION

Multiple sclerosis (MS) is a progressive neurological condition of the central nervous system characterized by inflammation, demyelination, and neurodegeneration. Symptoms experienced by people with MS are varied and differ between individuals. Commonly reported symptoms include, but are not limited to, reduced mobility, fatigue, difficulty with performing activities of daily living, and reduced community participation (1–3). These symptoms are often associated with barriers to engaging in exercise and physical activity within the home and community (4, 5). For purposes of this paper, physical activity is defined as "any bodily movement produced by skeletal muscles that results in energy expenditure" [(6), p. 126] and would include domestic, occupational, and sports-related activities. Exercise is defined as "a subset of physical activity that is planned, structured and repetitive" [(6), p. 126].

In the absence of a cure and with the limitations of disease-modifying therapies to stem disability accrual (7), exercise and physical activity remain a key strategy to manage the symptoms and consequences of MS. There is strong evidence that consistently demonstrates the safety (8, 9) and beneficial effects of exercise and physical activity (8). Examples include increased strength (8, 10), balance (8, 10), mood (8), mobility (8, 11), quality of life (8, 10, 12, 13), and fatigue (14). These effects have been shown to help people with MS manage MS symptoms and cope over time with the condition (15). However, despite the well-rehearsed safety and beneficial effects of exercise and physical activity, people with MS report lower levels of physical activity (16) and are reported as being sedentary (17–19).

Many different approaches have been developed to encourage more physical activity such as, "Blue prescription" (a physiotherapy approach designed to enhance adherence with physical activity in MS) (20–22), behavioral approaches (23–25), and self-management strategies (26). Although these have shown some promise in clinical trials, they have had limited impact on sustaining physical activity levels in people with MS. This highlights a potential mismatch between the evidence base and the reality of implementation for people with MS and the health (and other) professionals who work with them. As such, there is a need to reconsider the development of programmes and strategies to not only increase but also sustain physical activity levels in people with MS. That is, creating space for a multimodal approach that on one hand understands and addresses the drivers of physical activity from the lived experience while taking into account the framework and theoretical lens through which health professionals work.

Therefore, the aim of this paper is to demonstrate the interplay of a theoretical model widely used in clinical practice to gain further insight into exercise and physical activity. The authors draw on a series of studies (15, 27, 28) carried out sequentially using a mixed methods approach to provide additional insight into the lived experiences of people with MS and using the International Classification of Functioning, Disability, and Health (ICF) (29) as a conduit to discuss the key factors that drive decision making around exercise and physical activity in community-dwelling people with MS and its implications for health professionals. Following a brief overview of the ICF, three studies unpacking the meaning of exercise and physical activity will be summarized, followed by a discussion of the key findings framed within the ICF.

THE CLINICAL UTILITY OF THE ICF

The ICF is a global measure that is used to understand the health and health-related status of an individual (see **Figure 1**) (29). It consists of two key areas, namely, functioning and disability, and contextual factors. Functioning and disability include three domains, namely, body functions and structures, activity, and participation. The contextual factors include environmental and personal factors. This model recognizes the dynamic interactions that exist between the different domains of the ICF; for example, the influence the contextual factors (environmental and personal) might have on the outcomes of an intervention (5, 30, 31).

The ICF is underpinned by the principles of the biopsychosocial approach to understanding disability. Within this context, the ICF views disability as an interaction between the biological and social aspects of life (29). It is widely used to provide a common language amongst clinicians, researchers, and people with disability, including people with MS, to describe disability and contextual factors that might have an impact on their lives (30, 32–35). Therefore, given the ethos of the ICF, which focuses on bridging the clinical and experiential gap, it is used in this study as a useful tool through which physical activity can be examined.

THE MEANING OF EXERCISE AND PHYSICAL ACTIVITY: PERSPECTIVE MATTERS

Unpacking the meaning of exercise and physical activity is a complex issue that lends itself to an examination from different perspectives.

A three-phase mixed methods sequential explanatory design was used to determine the meanings people with MS ascribe to exercise and physical activity and its clinical implications (15, 27, 28). These studies have been published elsewhere but will be summarized here.

PHASE 1: A DAY IN THE LIFE OF PEOPLE WITH MS: THE DELPHI METHOD

A four-round Delphi questionnaire scoped and determined consensus of priorities for the top 10 exercise and physical activities and the reasons people with MS were engaged in these activities (27).

A purposive sample was recruited *via* a series of targeted strategies aimed at people who had the ability to complete questionnaires, who were diagnosed with MS, and who were living in the community ($N = 101$). Data were analyzed using content analysis, descriptive statistics, and non-parametric tests.

Findings from this study provided a snapshot view of exercise and physical activity. **Table 1** shows the top 10 prioritized exercise and physical activity practices and the reasons people with MS ($N = 70$) engaged in these activities. The consensus was achieved for the exercise and physical activities using Kendall's coefficient of concordance (36) ($W = 0.744$, $p < 0.0001$) and for the reasons they engaged in exercise and physical activity ($W = 0.723$, $p < 0.0001$). Overall, the exercise and physical activity practices and the reasons people with MS engaged in exercise and physical activity were diverse and highlighted the physical, psychological, and social benefits. Results indicated that unstructured activities

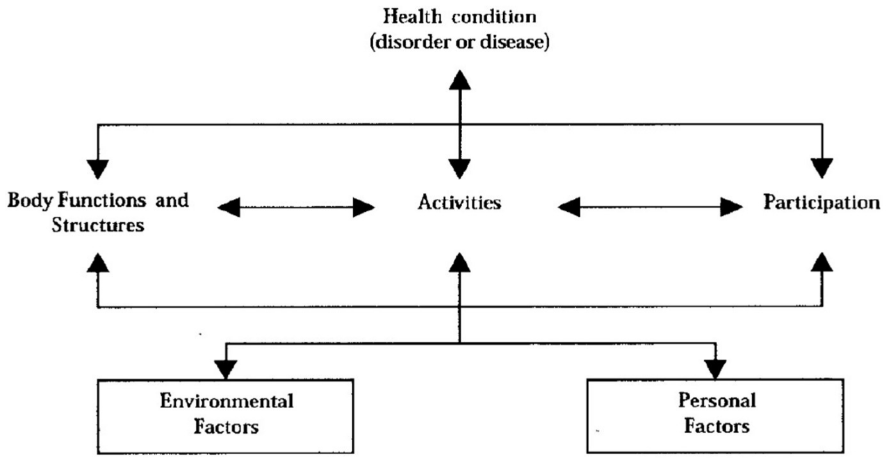

FIGURE 1 | Representation of the International Classification of Functioning, Disability, and Health (29).

TABLE 1 | Prioritised exercise and physical activity and reasons why people with MS engage in exercise and physical activity.

Rank	Exercise and physical activity priorities	Reasons why people with MS engage in exercise and physical activity
1	Self-care activities (e.g., shaving, shower, washing and dressing, cleaning teeth)	Improve MS symptoms (e.g., to improve or maintain strength, reduce pain, reduce spasms)
2	Every day activities (e.g., transferring, standing, pushing wheelchair or walking, climbing stairs)	Necessity (e.g., part of daily activities, activities that must be done)
3	Domestic activities (e.g., cooking, shopping, housework, laundry)	To keep active (e.g., to keep mind and body active, to maintain function and keep transferring)
4	Transportation (e.g., using public (bus/taxi) or personal transportation)	Mobility (e.g., to keep walking)
5	Leisure activities (e.g., gardening, dancing, sport, visiting friends)	Living with MS (e.g., fear of deterioration, combat MS)
6	Family Roles (e.g., spending time with family, looking after or playing with children/grandchildren)	Self-reliance (e.g., to maintain ones' independence and choice)
7	Stretches (e.g., activities done to stretch the upper and lower limbs)	Emotional wellbeing (e.g., pleasure, improve mood, reduce stress)
8	Physiotherapy (e.g., activities done with or by a physiotherapist)	Self-esteem (e.g., to manage weight, sense of achievement, self-worth)
9	Activities without weights (e.g., upper and lower limb exercises, bed and chair exercises)	Flexibility (e.g., to maintain or improve flexibility)
10	Technology (e.g., computer, exercise DVD, phone, Wii)	Social reasons (e.g., social, feel connected, be a part of community)

that focused on maintaining everyday function were prioritized and had a significant impact on the identity of people with MS.

PHASE 2: THE MEANING OF EXERCISE AND PHYSICAL ACTIVITY IN PEOPLE WITH MS: INTERVIEW METHOD

The meaning of exercise and physical activity was explored using in-depth semistructured, face-to-face interviews ($N = 16$; 12 women and four men). Data were analyzed using framework analysis (37). Details of the methods have been published elsewhere (15). To summarize the findings, five major themes were developed, namely, type of movement, impact of exercise and physical activity, "it changes", sense of loss, and coping with MS. **Table 2** provides a brief description of each theme. Overall, the study highlights views from the experiential perspective suggesting that people with MS took a multidimensional view of exercise and physical activity. This view of exercise and physical activity extended beyond movement; it was about using these activities as a way to cope, preserve their identity, and live life with a progressive neurological condition. Nonetheless, it was the contextual factors, such as, sense of loss and the fluctuating nature of priorities, energy demands, and choice that were dominant influences that dictated engagement or participation in exercise and physical activity.

PHASE 3: PERCEPTIONS OF PHYSIOTHERAPISTS ABOUT EXERCISE AND PHYSICAL ACTIVITY: FOCUS GROUP METHOD

Three focus groups were used to explore the understandings of physiotherapists about exercise and physical activity in

TABLE 2 | Major themes and subthemes for the meaning of exercise and physical activity.

Main theme	Sub-theme	Description of the theme	Examples of quotation
A type of movement	• Exercise as specific and organized movement • Physical activity as part of daily routine • Relevant for life with MS	Exercise and physical activity were about movement. However, the nature and purpose of each movement was different.	• Sue:…*exercise is something that, in a funny kind of way.. doesn't form part of one's kind of routine… something that you make separate..time for… amm.. but so, it's so… slightly more in isolation, to all the things that you would do...* (Sue, 50 years, F, severely affected, line 91–94) • *James:…physical activity I need to move from here to there and use whatever I can do, to stand up, walk, move upstairs, that, that's all physical activity to me* (James, 53 years, M, moderately affected, line 117–120)
Impact of exercise and physical activity	• Physical impact • Psychological impact • Social impact	Participants described the positive benefits of exercise and physical activity on the physical, psychological and social aspects of life.	*… feel good factor. …I don't know what it is in your body that when you exercise it sort of seems to release all these bits and pieces and it makes you feel better* (Linda, 71 years, F, moderately affected, *line 142–158).*
It changes	• Reflections on the past and ever changing present • Uncertain future • The influence on priorities	It changes illustrate that the meaning of exercise and physical activity was contextualized to the progressive nature of MS and personal life situation.	*..you know things change so obviously …amm.. exercise will change.. depending on your.. circumstances, ammm… as you get older you do a different type of.. I mean, I'm speaking for myself.. I do a different type of exercise than I would of.. I also do different things now that.. I've got MS, than before I had MS. (Pam, 65 years, F, Moderately affected, line 58–66)*
Sense of loss	• Loss of activity (loss of independence) • Loss of employment • Compromise and reconciliation	Participants described multiple losses. The ability to undertake certain physical activity was associated with significant loss in different areas of life such as hobbies and employment. Compromise with certain activities and a sense of reconciliation about what was loss was seen in the excepts.	*.. in the past I use to love walking and would walk for hours and this is a great loss to me..I realize I can't really do the walking I use to do.. (Bev, 55 years, F, moderately affected, line 90–93)*
Coping with MS	• Normalcy • Control over physical symptoms • Exercise and physical activity frames the week • Support • This is me"- identity	Participants used exercise and physical activity as a way to cope, shape and preserve their sense of self.	*…. Classes do sort of, give a framework to my week.. I would think oh its x day, so x day this time I will be going to Pilates class or there would be y day.. and, if I go to the Physio sort of session.. I would go to that.. because I am not working now if I didn't have that structure to my day… ammm… I could see the whole thing sort of falling apart! (Bev, 55 years, F, moderately affected, line 142–149)*

light of the Delphi results and their relevance to clinical practice (28). Physiotherapists ($N = 14$; 12 women/2 men) with experience working with people with MS in the community were included. The focus groups were analyzed using the principles of framework analysis (37). Four themes were developed, namely, blurred terminologies, influencing factors for the meaning of exercise and physical activity, when professional expertise meets experiential expertise, and the resolve. **Table 3** shows the themes and a brief description of each theme. Overall, the findings highlight the perspective of professionals that was largely shaped by training and models of practice. Physiotherapists expressed that the use of exercise is embedded into clinical practice, but physical whilst activity considered is less routine in clinical practice.

THE INTERPLAY OF THEORY AND PHYSICAL ACTIVITY USING THE ICF

To explore the interplay of physical activity from the lived experience and the experience of professionals and how they interact with the theory, the key findings were mapped onto the domains of the ICF (29). The findings from the perspectives of people with MS (Phase 1 and 2) (see **Figure 2**) were mapped separately to those of the health professionals (see **Figure 3**).

Conceptually, the diagram highlights that the exercise and physical activity practices, and the meanings people with MS ascribed to exercise and physical activity fit within the ICF model multiple times across all of the domains of function, disability, and contextual factors. This supports the applicability of the ICF

TABLE 3 | Major themes and subthemes for perceptions of physiotherapists of exercise and physical activity.

Main theme	Sub-theme	Description of themes	Examples of quotation
Blurred terminologies	• Attributes of exercise and physical activity • "I kind of don't agree with my own definition"	Participants described the sense that exercise and physical activity were intricately linked. Discussions revealed attitudes toward exercise and physical activity and conflicts with the definitions used.	*..Things like walking the dog, walking to the shops, carrying the shopping. As its maybe a less intensive form of exercise (FG1, 270–274)*
Influencing factors for the meaning of exercise and physical activity	• Training vs. pragmatism in the community • External factors: Use of language, government initiatives coupled with lack of resources.	Participants discussed a number of factors that influenced the meaning of exercise and physical activity. These were described based on their training and other external factors.	*..right so we work in the NHS – you can't keep people … we're not allowed, and we can't see people every week for exercise or stretches. And mainly from a resource point of view initially, but also in terms of sort of the self- management, you know the expert patient, you know facilitating patients to manage their conditions … I think you then end up looking at exercise in a very different sort of way, cos it's not something that they're coming to you for – you are trying to encourage them to take on board the principles and then do it in their everyday life. (FG1, 462–475)*
When professional expertise meets experiential expertise	• Creation of inner tensions • Making sense of Delphi Results.	This theme reflects some of the attitudes within the study when the prioritized exercise and physical activities and the reasons why people with MS engaged in exercise and physical activity were viewed. The priorities of people with MS challenged physiotherapist understanding about the therapeutic approach used in the management of MS in the community.	*It seems ridiculous but I suppose... it wasn't how I was thinking, more than I'm surprised. I was kind of... because of the exercise thing that I conceded in my head, it was more like you know what's the most popular way to exercise rather than... more just activity. (FG2, L, 813–818)*
The resolve	• Positive reinforcement of current practice • Re-evaluation of current practice.	Through discussions, negotiations and deliberations within the group Physiotherapist attitude shifted during the focus group as they reflected on their own practice.	*See I think that one*; activities due to family roles*, I don't really address, and I think that's probably 'cos I don't have children and my family don't live nearby. So I think that's probably something that is good to have brought up.. (FG2, 921–936)*

to the lived experience of people with MS in relation to exercise and physical activity.

The findings from the perspective of professionals were also mapped onto the ICF conceptually to ascertain how their views about exercise and physical activity fit within this model (see **Figure 3**). The representation of the perspective of physiotherapists highlights less focus on the participation and contextual factors domains. Of note, there were double the number of factors within the function and disability domain compared with the contextual domain. Indeed, the contextual factors reported by physiotherapists were less than half expressed by people with MS.

Both perspectives were then merged to compare and contrast the views of people with MS and physiotherapists (see **Figure 4**). It illustrates each domain and highlights that certain domains of the ICF had a greater influence on how people with MS and physiotherapists ascribed meaning to exercise and physical activity. These influences will be discussed to highlight areas of overlap and areas of dominance.

The findings from the experiential perspective demonstrated that people with MS adopted a participatory mind set, as their discussions focused on social and leisure-type activities as well as the use of technologies that enabled access to these activities. As such, they were more likely to engage in activities that connected with other people. This was in contrast to physiotherapists whose concerns revolved around whether the activities people with MS prioritized would have a direct impact on their physical performance. These findings would suggest that for physiotherapists, greater weighting and higher priority were given to the body structure, function, and activity domains rather than participation domains of the ICF in ascribing meaning to exercise and physical activity.

In contrast, the functioning and disability domains reflected a different weight of influence for people with MS compared

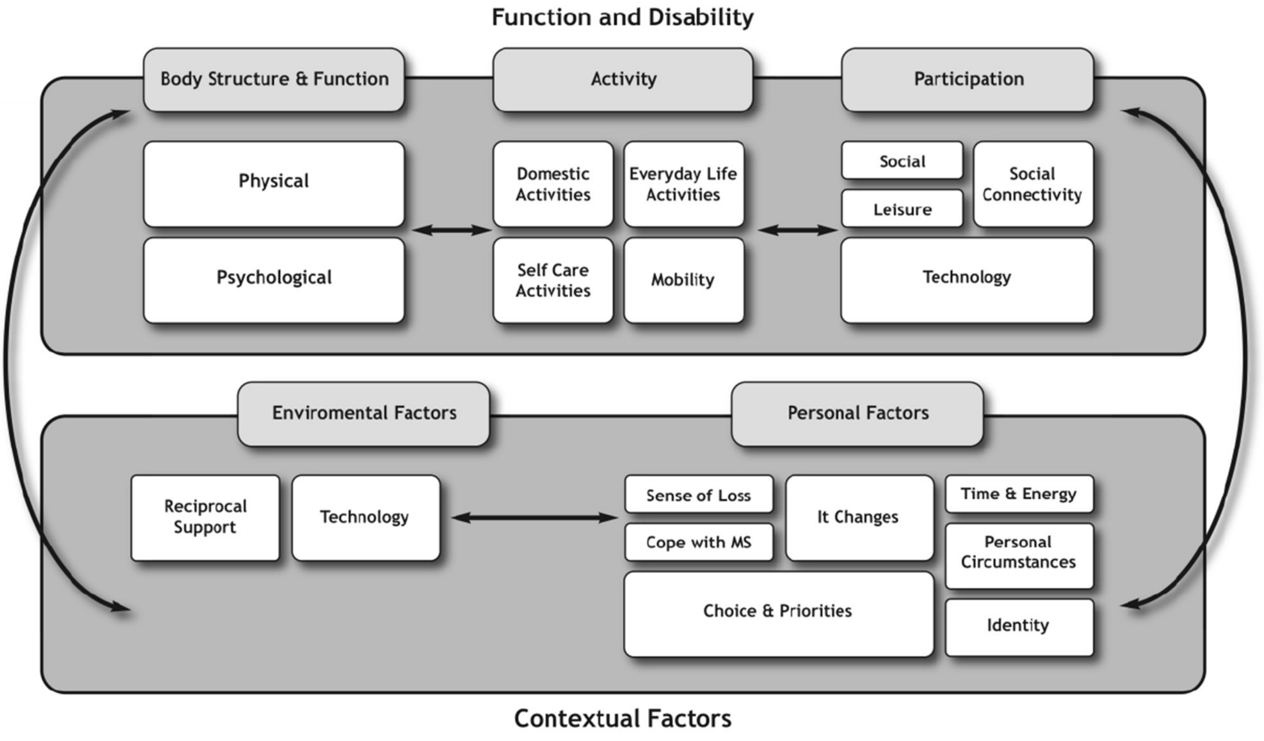

FIGURE 2 | Representation of the experiential (people with MS) perspective in relation to the ICF.

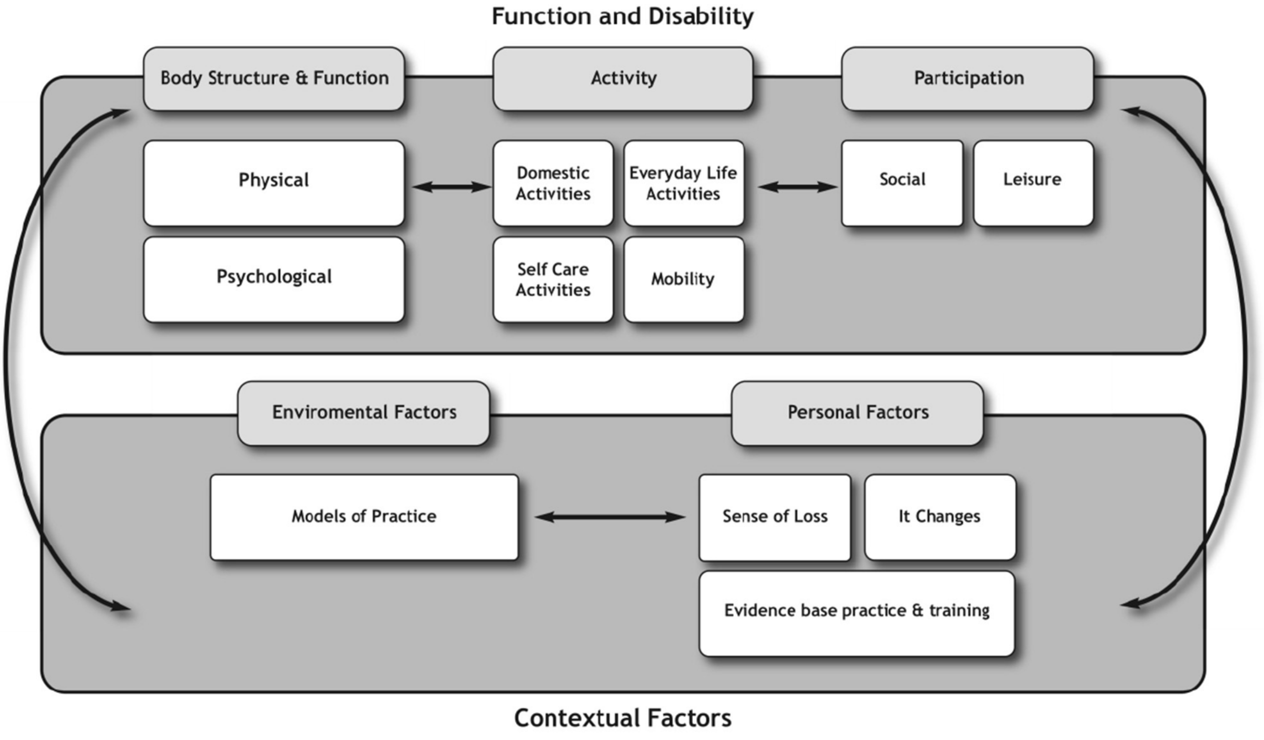

FIGURE 3 | Representation of the professional (physiotherapists) perspective in relation to the ICF.

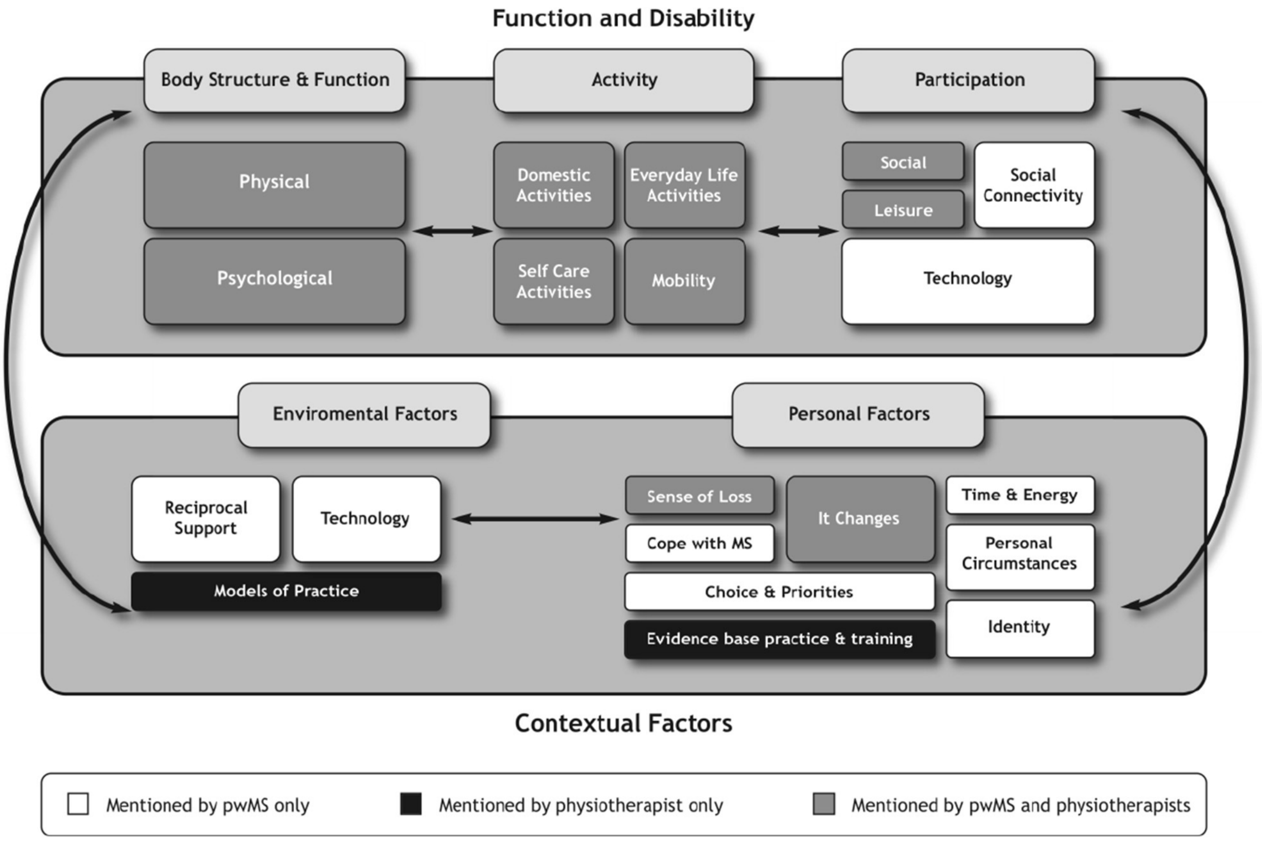

FIGURE 4 | Representation of the experiential and professional perspective in relation to the ICF.

with physiotherapists. For example, the perceptions of people with MS about exercise and physical activity were dynamic and diverse and could be reflected across all the domains of the ICF. This was exemplified by the inclusion of certain categories such as transportation and activities involving technology, which were prioritized by people with MS in the Delphi study (27). The inclusion of technology was interesting and traversed the participation and environmental domains. For example, people with MS described technology as a form of activity to facilitate higher education, as well as the use of technology as a communicative device necessary for organizing daily routines. These were not mentioned by physiotherapists, and signify the expansive views of people with MS in relation to exercise and physical activity.

Furthermore, the findings from the interviews revealed that in ascribing meaning to exercise and physical activity, people with MS were influenced predominantly by the participation and contextual factors domains; more specifically the personal factors. These contextual factors, which included the environmental and, specifically, the personal factors, shaped the perspectives of people with MS (see **Figure 2**), for example, "coping with MS," "identity," "energy demands and availability," "time constraints," "personal choice and priorities." These findings concur with other researchers who have also identified that contextual factors play a significant role in influencing the other domains such as functioning and disability in people with MS as well as in people living with other forms of disability (31, 33, 35, 38). Therefore, understanding the influence contextual factors play in people with MS is important especially to health professionals who use exercise and physical activity as treatment strategies. Lack of understanding and insight into these contextual issues render people with MS seemingly inactive to the view of professionals, whereas the findings from the experiential perspective portray a different picture where people with MS are active on other priorities in other contexts.

The responses of physiotherapists did not fit neatly into the ICF framework. For example, physiotherapists shared aspects of the themes "*sense of loss" and "it changes,*" which represents the personal factors of people with MS. This finding suggests that physiotherapists do consider some aspects of the personal factors identified by people with MS. However, physiotherapists did not make the link as to how these personal factors might influence engagement in exercise and physical activity beyond the physical aspects of the life of individuals. In addition, the findings from the perspectives of physiotherapists highlight that their views about exercise and physical activity were also shaped by their own contextual factors, which were external to people with MS but influenced decisions around their management in the community. These factors included their professional knowledge based on evidence-based practice and training (personal factors)

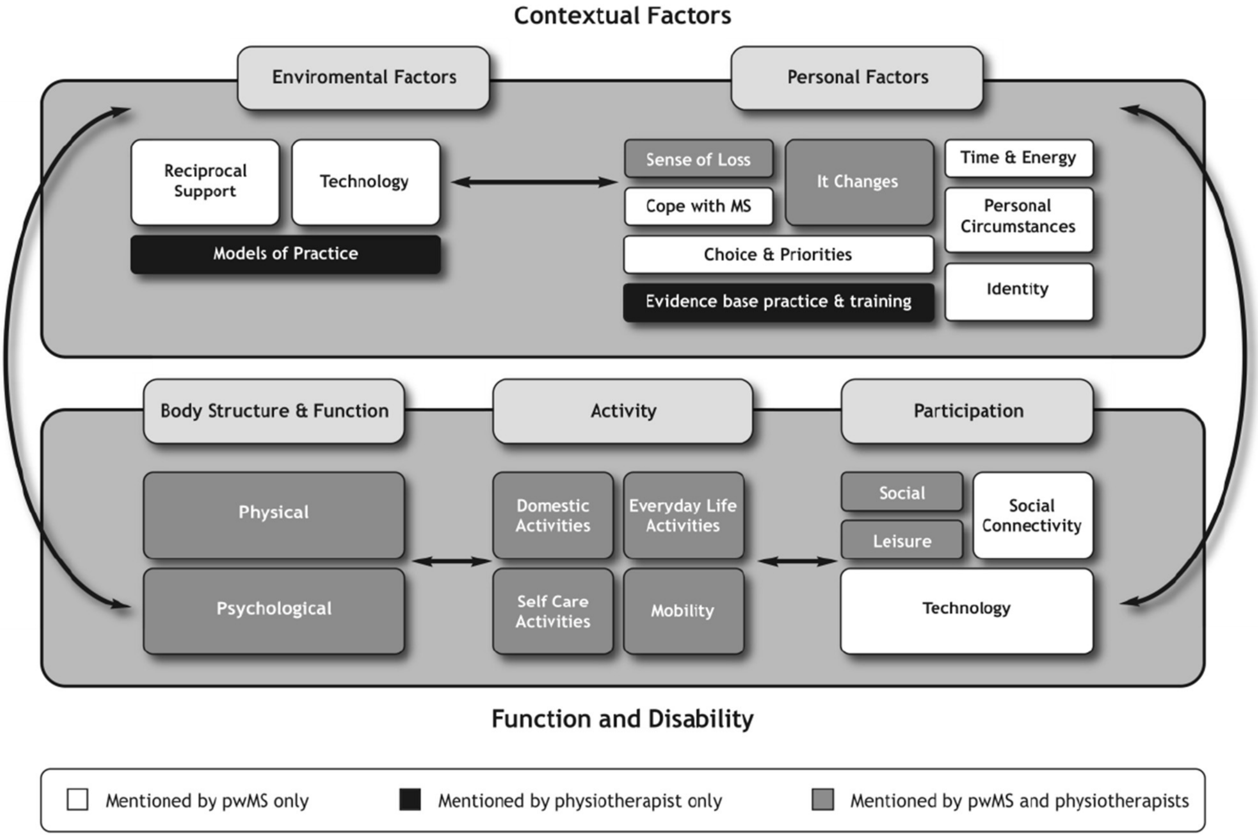

FIGURE 5 | Reorientation of the ICF based on the experiential and perspective of the professionals of exercise and physical activity.

as well as models of practice, which could be represented under environmental factors.

The current interpretation of the ICF implies that the contextual factors interact with the functional and disability domains (29). Although this is true, this study extended this view to also suggest that for community-dwelling people with MS, the contextual factors did not only influence the functional and disability domains but dictated what happened at the functional and disability domains. Indeed, the orientation of priority was challenged by people with MS. Having considered the views put forward by people with MS, the importance of how these views were expressed, and the heavier weight attributed by the contextual factors, the authors reconsidered the orientation of the ICF by 180°. This flip suggests that the contextual factors played a more major role than previously thought in relation to the exercise and physical activity practices, and the meanings people with MS ascribed to these practices (see **Figure 5**).

IMPLICATIONS FOR FUTURE THINKING ON PHYSICAL ACTIVITY IN PEOPLE WITH MS

Reconceptualising the interactions between the contextual factors and the function and disability domains, not only as influences to be considered but also as factors having the capacity to dictate decisions about exercise and physical activity, should be central to the thinking behind engagement strategies in community-dwelling people with MS. As such, physiotherapists and other exercise professionals working in the community should give more focused attention to these domains when designing and implementing rehabilitation strategies or programs for people with MS living in the community as a way to engage and sustain exercise and physical activity in this population.

This study provided some insights about exercise and physical activity based on two sources of information, one extracted from the experiential perspective of people with MS and the other from the perspective of professionals. Exploring both sources of information suggests a "rethink" about how exercise and physical activity are viewed by health professionals and points toward taking a more person-centered approach to reflect the preferences and priorities of community-dwelling people with MS.

The key findings from the experiential perspective have not only identified the exercise and physical activity preferences and priorities of people with MS but also what these activities mean to people with MS. Having an understanding of these activities and their meanings provides some insight into the way health professionals, specifically, physiotherapists might approach exercise and physical activity in people with MS living in the community. For example, people with MS preferred engaging in exercise and physical activity practices that they valued and considered meaningful for living life with MS. This suggests that exercise and physical activity for community-dwelling people with MS were more than managing MS symptoms and also about the importance of participating in life activities, how they coped with life and maintained a sense of self. Therefore, it is now time to create more opportunities and design interventions that reflect the participatory aspect of exercise and physical activity and develop tools to monitor such interventions with a participatory focus.

STRENGTHS AND LIMITATIONS

Using the key findings from a mixed methods study, this paper provided some insights into the intricacies associated with exercise and physical activity from the perspectives of people with MS and its clinical applicability to health care professionals. It also highlights the types of personal factors and their relevance to dictate and influence engagement in physical activity in people with MS. However, the findings must be examined, reflected on, and interpreted within the context and rigor of each study. As such, the findings might not be generalisable beyond the participants and context of the studies highlighted in this paper. Nonetheless, further research could explore the theoretical underpinnings and concepts highlighted in this study in other long-term conditions and contexts.

CONCLUSIONS

This paper demonstrates the interplay between theory and physical activity in people with MS using the ICF model to guide discussions. The model illustrates the interaction of the ICF domains in relation to the meanings ascribed to exercise and physical activity based on the perspectives of people with MS and physiotherapists. It highlights that although people with MS were predominately influenced by participation and personal factors, physiotherapists were predominately influenced by the function and disability domains, albeit with less reference to participation. In addition, this paper adds to the existing evidence in relation to exercise and physical activity and provides evidence that the perception of exercise and physical activity in people with MS is not static and limited to any one domain within the ICF model. Instead, it highlights a complex concept, which is dynamic in nature, traversing between functioning and disability and contextual factors (personal and environmental) with personal factors having a greater influence on decisions made about exercise and physical activity in people with MS.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/supplementary material, further inquiries can be directed to the corresponding author/s.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by School of Health Sciences and Social Care, Brunel University. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

All authors read and contributed to the manuscript development and revision of this paper.

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Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers
in Rehabilitation Sciences

PERSPECTIVE
published: 21 October 2021
doi: 10.3389/fresc.2021.705474

Proposing a Core Outcome Set for Physical Activity and Exercise Interventions in People With Rare Neurological Conditions

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Specialty section:
This article was submitted to Disability, Rehabilitation, and Inclusion, a section of the journal Frontiers in Rehabilitation Sciences

Received: *05 May 2021*
Accepted: *23 September 2021*
Published: *21 October 2021*

Citation:
Ramdharry G, Buscemi V, Boaz A, Dawes H, Jaki T, Jones F, Marsden J, Paul L, Playle R, Randell E, Robling M, Rochester L and Busse M (2021) Proposing a Core Outcome Set for Physical Activity and Exercise Interventions in People With Rare Neurological Conditions. Front. Rehabilit. Sci. 2:705474. doi: 10.3389/fresc.2021.705474

Rare neurological conditions (RNCs) encompass a variety of diseases that differ in progression and symptoms but typically include muscle weakness, sensory and balance impairment and difficulty with coordinating voluntary movement. This can limit overall physical activity, so interventions to address this are recommended. The aim of this study was to agree a core outcome measurement set for physical activity interventions in people living with RNCs. We followed established guidelines to develop core outcome sets. Broad ranging discussions in a series of stakeholder workshops led to the consensus that (1) physical well-being; (2) psychological well-being and (3) participation in day-to-day activities should be evaluated in interventions. Recommendations were further informed by a scoping review of physical activity interventions for people living with RNCs. Nearly 200 outcome measures were identified from the review with a specific focus on activities or functions (e.g, on lower limb function, ability to perform daily tasks) but limited consideration of participation based outcomes (e.g., social interaction, work and leisure). Follow on searches identified two instruments that matched the priority areas: the Oxford Participation and Activities Questionnaire and the Sources of Self-Efficacy for Physical Activity. We propose these scales as measures to assess outcomes that are particularly relevant to assess when evaluating physical activity interventions mong people with RNCs. Validation work across rare neurological conditions is now required to inform application of this core outcome set in future clinical trials to facilitate syntheses of results and meta-analyses.

Keywords: physical activity, neuromuscular disease, motor neurone disease, Huntington's disease, inherited ataxias, hereditary spastic paraplegia, parkinsonism, outcome measurement instruments

INTRODUCTION

Rare neurological conditions (RNCs), where cases are ≤40 per 100,000 population (1), collectively incur a significant cost burden to healthcare, social care services and informal care (2). Despite variability across conditions, many of these conditions will share symptoms and signs at the level of body function, activities and participation (3). As such, common approaches to improve fitness (e.g., cardiovascular and strength training), activity (e.g., balance and gait training) and participation levels (e.g., supported self-management) are often implemented in clinical practice (4–8).

Physical activity is any bodily movement produced by the muscles that require us to expend energy and can include structured exercise, active transportation, household chores, and activity during work, play and recreation (9). Whilst trials of physical activity interventions in RNCs highlight the potential of physical activity interventions to improve fitness and function (10–13) they are often small studies that fail to influence clinical practice. There are a variety of factors that limit the impact of these trials, not least the selection of outcome measures. Measurement constructs may vary from physiological measures (e.g., strength and fitness), to functional assessments (e.g., walking speed, climbing stairs) or quality of life and well-being outcomes and do not typically take into account patient preferences (14). If we are to ensure that research is relevant and able to influence clinical practice and future research, we need to ensure the use (and reporting) of standardized, relevant outcome measures within the field that are applicable to people living with the conditions (15, 16). Importantly, it should not be assumed that measurement should be restricted to the agreed core outcomes but rather that these outcomes should always be gathered and reported to facilitate evidence synthesis across relevant trials and studies. Core outcome sets have been agreed for specific target conditions for example cancer, rheumatology and chronic pain as well as for specific care pathways for example maternity care (17, 18).

Core outcome sets have also been proposed for people with neurologic conditions (19), including adults with dementia (20). However, these core sets have not been tailored to physical activity interventions for people living with RNCs. Recommending measures would not only help to bring consistency in reporting, allowing comparisons or meta-analysis of future studies, but also ensure responses are measured of constructs important to people living with RNCs. This study focuses on the development of an agreed standardized set of outcomes termed a "core outcome set" (21) that should be at a minimum measured and reported in trials of physical activity interventions in people living with RNCs.

METHOD

The study followed the guidelines of the COSMIN (COnsensus-based Standards for the selection of health Measurement INstruments) and COMET (Core Outcome Measures in Effectiveness Trials) initiatives. A four step approach was used to select outcome measurement instruments recommended within core outcome sets (21). The activities relevant to each step within the process are described in detail below.

We focused on groups of progressive RNCs, namely neuromuscular diseases, Ataxias, Huntington's Disease (HD), Atypical Parkinson Diseases (AP), including Progressive Supranuclear Palsy, Multiple Systems Atrophy, Corticobasal Degeneration, Motor Neuron Diseases (MND) and Hereditary Spastic Paraparesis (HSP). These conditions affect ∼2–10 per 100,000 in the general population, collectively leading to limited mobility and poor balance for many individuals. People with neuromuscular diseases and MND experience profound weakness and muscle atrophy. People with Ataxia, HD, AP, and HSP experience difficulty controlling movement, with some muscle weakness and variable cognitive impairment. Many people across the conditions also experience pain, joint deformity, fatigue and depression which impacts on their ability to participate in routine activities of daily living.

Step One: Conceptual Considerations

People living with RNCs, carers of people with RNCs and representatives from five collaborating support groups and charities, namely the Muscular Dystrophy Association, Ataxia UK, HSP support group, PSP Association, HD Association of England and Wales were invited to join a stakeholder group. They attended an initial workshop (Workshop 1) to define conceptual considerations in relation to the physical activity interventions and outcomes in our target population, namely, people living with RNCs.

This was followed by a second stakeholder workshop (Workshop 2) with people living with RNC and the relevant charity representatives. They worked together to (a) explore issues and experiences relating to physical activity in the face of living with a RNC and (b) identify and priorities key constructs and domains of importance that would need to be measured when evaluating a physical activity intervention. Representatives from RNC charities were asked to gather views from their members living with RNCs prior to the meeting through their communication channels, e.g., surveys and social media platforms.

Step Two: Finding Existing Outcome Measurement Instruments

We conducted a scoping review of systematic reviews published between January 2008 and December 2018 to identify outcome measures used to measure efficacy of any type of physical activity intervention for adults with neuromuscular diseases, motor neurone disease (MND), HD, PSP, multiple system atrophy (MSA), inherited ataxias and HSP (Open Science Framework registration: https://osf.io/4cr32/). The research team, experts in this field, were aware that little research into physical activity had taken place until the early 2000s and reviews came later, hence the 10-year window. Studies were included if participants were adults and if the reviews reported at least one outcome measure to evaluate the efficacy of the physical activity intervention at either the body structure/function, activity and/or participation levels, according to the International Classification of Functioning, Disability and Health (ICF) (20). Constructs and domains of importance identified during Workshop 2, were matched with outcome measures identified in the scoping review. Where no measures matched the identified, domains, additional literature searches were done using the domain descriptions as search terms. Additional criteria for selection were use in RNC or other neurological diseases. Two of the descriptors, motivation and confidence, relate strongly to self-efficacy so this was also added as a search term. Elicitation of stakeholder opinions during a series of virtual meetings was undertaken to supplement this process and to identify proposed outcome measures that were consistent with the domains of importance identified by the stakeholder group.

Step Three: Quality and Feasibility Assessment of Outcome Measurement Instruments

Outcome measurement instruments that are included in a core outcome set should ideally be reliable and valid for use in the target populations (22). Feasibility of use is a further consideration. The rarity of the diseases being studied meant that the psychometric properties of the measurement tools we identified had not been examined in these conditions. We thus considered the psychometric properties of the tools as applied to more common long term, neurological conditions where there were indications of some common impairments. The evidence for each was collated for presentation at step four.

Step Four: Reaching Consensus on the Proposed Core Outcome Set

The outcome measure instruments under scrutiny matching the agreed domains of importance were examined by individual researchers then presented to the wider research team for technical discussions of the psychometric properties through a series of video meetings. Following the video meetings, lists of items assessed within each instrument were sent to the stakeholder group via e-mail to elicit further reflection on their relevance to the constructs of importance. A final face to face stakeholder workshop (workshop 3) and consensus procedure was undertaken to agree on the instruments for each outcome to be recommended for inclusion in the core outcome set.

RESULTS

Step One: Conceptual Considerations

People living with RNCs (workshop 1 $N = 5$; workshop 2 $N = 3$), carers (workshop 1 $N = 1$) and charity representatives (workshop 1 $N = 5$; workshop 2 $N = 5$) considered it important that measurement tools were able to detect outcomes across domains of (A) function and well-being and (B) participation in activities. In terms of (A), staying well, ensuring good sleep and maintaining positive mood were of highest priority whilst in relation to (B), the ability and confidence to take control and make choices along with normalization of participation and social engagement were important. Through further discussion, the stakeholder groups agreed that these aspects were well-centered around (i) physical well-being; (ii) psychological well-being and (iii) participation in day-to-day activities as the primary domains of meaningful importance. Relevant constructs within the physical domain were physical function and independence. Constructs in the psychological well-being domain were emotional well-being, mood, enjoyment, motivation for physical activity and confidence, whilst leisure activities, work and activity that matters (personal choice) were constructs of importance within the domain of participation.

Step Two: Finding Existing Outcome Measurement Instruments

Database searches identified 5,435 articles, and, after removing duplicates, 4,433 were screened by titles and abstracts, leaving 62 articles for full-text eligibility assessment. They were screened and 27 were included in the scoping review (4, 5, 8, 12, 13, 23–44). The results of the scoping review will be presented in detail separately. Nearly 200 outcome measures assessing outcomes of structured physical activity interventions (**Table 1**) were identified within these 27 articles. Dosage, intensity and duration of training regimes were highly variable but typically involved strength training, aerobic and respiratory, functional training and combined programs with very few focusing on physical activity behavior change.

We mapped each outcome to the World Health Organisation International Classification of Function (ICF) domains (20) (see **Table 2**). The majority were related to function and activity. Outcomes reflective of both body structure impairments and participation were less frequently reported. Two domains were categorized as "Other" (e.g., Goal attainment score), and "Disease-specific" questionnaires (e.g., Unified Huntington's Disease Rating Scale or Scale for the Assessment and Rating of Ataxia).

Eleven reviews utilized disease-specific outcome measures, while in six reviews measures were not able to be represented within the ICF domains (i.e., in the "Other" category). Most studies ($n = 17$) included outcomes that were representative of three to five different domains. Notably, there was no evidence of stakeholder engagement or involvement of people with the condition being investigated in the selection of measures used as primary outcomes.

Constructs relevant to the *physical and psychological well-being*, and *participation in day-to-day activities* domains were cross-checked with the outcomes synthesized in the scoping review. No single outcome measurement instrument that addressed all three domains was identified. Measures were usually tailored to specific activities or functions (e.g., on lower limb function, ability to perform daily tasks). Alternative outcomes reflective of well-being were reviewed by stakeholders through a series of group discussions. None of these comprehensively matched the domains of importance identified in Step One.

Further literature searching resulted in the Oxford Participation and Activities Questionnaire (Ox-PAQ) (50, 51) being identified as an instrument that matched the majority of the constructs highlighted as relevant in

TABLE 1 | Characteristics (including outcome measures utilized) reported in studies included in scoping reviews of physical activity interventions in RNCs.

References	Research designs of included studies	Number of participants	Participant characteristics	Controls	Outcome measures
Cup et al. (24)	2 studies using pre-post design	17	Male/female: 7/10 Mean Age (Y): Study 1: 52.4 (range: 28–67) Study 2: 62.5 (range: 39–83)	No controls	Performance and satisfaction/Muscle strength/Grip force and pinch grip (Grippit)/Fine motor control (Purdue Pegboard), Range of Motion (goniometer). Activities of daily living (interview with ADL-Taxonomy); Life satisfaction (modified Life Satisfaction Checklist)
Habers and Takken (27)	2 RCTs, 1 non-RCT, 9 uncontrolled studies	161	Male, *N* = 57; Females, *N* = 104 Mean Age (Y): range 40–68	Not reported	Disease activity (e.g., serum levels of creatine kinase, aldolase, cytokines etc.)/Muscle strength/Aerobic fitness/Functional performance/Functional capacity/Health status/Lung function/Muscle characteristics/Disease impact/Fatigue
Quinlivan et al. (13)	3 non-randomized studies	27	2 out of 3 studies reported gender: 9 males and 9 females in total. Mean Age (Y): range 32–61	Same training but in healthy controls, age- and sex-matched healthy, sedentary controls otherwise not specified	Borg rating of perceived exertion/VO2 max /HR/Superficial EMG for muscle activity and glucose and lactate blood levels/Serum creatine kinase /Respiratory gas exchange/Cardiac output and serum samples for lactate and glucose were measured/Needle muscle biopsy of vastus/Respiratory gas exchange data were collected/VO2 peak, and gross mechanical efficiency during the constant workload test/HR using 12 lead ECG tracing/Capillary blood samples lactate and ammonia
Ydemann et al. (28)	1 Pre-post cohort, 4 RCTs, 1 prospective cohort, 1 quasi-RCT, 1 descriptive study	757	Not reported	Standard medical therapy, usual care, daily interruption of sedation only, general physiotherapy alone	6 MWD/MIP/Isometric quadriceps force/Subjective feeling of functional well-being/Time in bed/ICU stay/Hospital stay/Duration of delirium/Ventilator-free days/Muscle fatigue and isotime dyspnoea/Atrophy/Weaning of atrophy (no further details provided)
Voet et al. (23)	4 RCTs and 1 quasi randomized study	170	20 adults with mitochondrial myopathy, diagnosed on the basis of clinical, familial and muscle biopsy data. 35 adults with myotonic dystrophy type 1, genetically confirmed. 36 adults with myotonic dystrophy (2 congenital form, 34 classical adult type), diagnosis not verified. 65 adults with FSHD, genetically confirmed. 9 adults with dermatomyositis and 5 adults with polymyositis.	Strength training vs. no training	Differences in Muscle strength (using dynamometer)/Quantitative Muscle Assessment fixed myometry testing system/Dynamic strength was evaluated using the one repetition maximum / Weight-lifting capacity/ Endurance time measured in a submaximal cycling test at a constant workload of 70%/ 6 MWT, VO2 max/ Maximum duration of contraction at 80% of MVIC on an isokinetic dynamometer/Sickness Impact Profile and the Symptom-Checklist /Nottingham Health Profile/ SF-36 Health Survey/ CIS-fatigue
Gianola et al. (25)	4 (3 controlled and 1 randomized clinical) trials. One study was excluded as included participants under 18 years old.	128	Mean Age of participants range from 22 to 48 years	No control/healthy control group/other interventions	Maximal voluntary isometric contraction /Maximal peak torque/Modified MRC/Six-Minute Bicycle Test/BORG/Six-minute walk test (m)/Timed-stands test/Timed up-and-go test/MVIC isokinetic torque/Test 80% MVC (sec)/Descending stairs/Climbing stairs/Standing up from a chair/Standing up from lying supine/Walking 6 min (comfortably)/Walking 50 m (fast) (sec)/CIS-fatigue/ICF functional dimensions
Narayanaswami et al. (26)	5 Class III studies	62	12 patients with Welander distal myopathy 9 ambulatory patients with LGMD2I and 9 healthy controls 11 men with BMD and 7 healthy men 8 patients with hIBM3 secondary to a defect in the MYH2 gene 6 patients with hIBM3 secondary to a defect in the MYH2 gene	Sedentary, age-matched controls	Maximal oxygen uptake/Maximal workload, and other patient-reported outcomes/Maximum workload/Muscle strength/Change in the expression of myosin isoforms on muscle biopsy

(Continued)

TABLE 1 | Continued

References	Research designs of included studies	Number of participants	Participant characteristics	Controls	Outcome measures
Khan and Amatya (32)	1 SRV, 1 RCT, 1 case-control, 5 prospective or retrospective cohort studies, 6 case series/reports	422	Not reported	Low-intensity home based program of maintenance exercises and education for self-management (30 min twice a week) (RCT) or healthy controls.	HRQoL/FIM/PIPPS/DASS/WHOQoL/LOS/Modified Barthel Index/MRS /HDS NHP/BI/ESS/HAS
Simatos Arsenault et al. (12)	1 single subject, 4 case reports, 1 quasi experimental design, 1 RCT	133	66 females vs. 67 males Mean Age (Y): 43.8	No exercise or lower intensity, home-based ambulatory exercise, otherwise not reported	FSS/Activity monitor/SF-36, FIS/Perceived mental functioning/Physical fitness (peak work levels, VO2 mL/min, mL/kg/min)/Ventilation/Isokinetic leg strength (total work capacity)/General mobility/Confidence in walking/Cardiorespiratory cycle ergometer test/Isokinetic muscle strength/Functional outcome of daily physical activity (RAM)/FIS (cognitive, physical, and social)/GBS disability score/HADS/RHS/QOL SF-36/MMT/WHOQOL-BREF/DASS-21/PIPP/Physical fitness (duration of exercise, distance walking, distance cycling, grip strength)/Pulmonary fitness (PEFR, FVC, FEV1)
Young et al. (29)	1 Randomized controlled single blind trial	29	Not reported	No strength training	Muscle strength voluntary contraction/Isokinetic knee torques/Timed functional activities
Sman et al. (5)	3 RCTs, 5 quasi-experimental (i.e., pre-post testing), 1 case report	134	Average age: 38 years old. 8 out of 9 studies reported gender: 52% were male, 48% female.	Where reported: controls underwent the same program, however, balance training was managed by a physiotherapist instead of a mechanical apparatus	Muscle strength (N or Kg)/Maximal voluntary isometric testing (Kg)/Isokinetic knee torque flex/extension/MVC/Endurance test at 80% MVC/Isokinetic muscle strength (Nm)/Medical research council scale (MRC)/BOT (balance) score/Power/Long jump (cm)/6 MWT/Walking ability (different parameters)/Functional activities (e.g., Chair raise)/CMTES/Phone FITT FDI/ROM/Tinetti Scale/Berg Balance scale/Physiological (BMI,FFM, Percent body fat, Serum myoglobin, RMS (µv)/Fatigue Severity Scale/Modified PCI/MHC/Myosin heavy chain/Cardiorespiratory cycle test/Mean blood CK/VAS/VO2 max/HR/Respiratory Borg Scale/METS/Fatigue Borg Scale
Corrado et al. (30)	4 RCTs and 1 Cohort	236	Not reported	No intervention	Quantitative neuromuscular assessment/Bioelectrical impedance analysis/6 MWT/MVC (myometer or isokynetic dynamometre)/Borg scale/Serum level of myoglobin/Surface electromyography techniques/Holter
Lui and Byl (33)	Prospective clinical studies (*N* = 2), RCTs (*N* = 2) and 1 SRV (Dalbello-Haas et al. (47), previous version of Dal Bello and Florence (4))	98 (including Drory et al. (45); Bello-Haas et al. (46)), excluding Dalbello-Haas et al. (47)	Not reported	Usual care/home exercise program without supervision/no exercise participation or usual activities	Norris ALS score strength/ALS-FRS strength MMT/FSS/FIM/FVC

(Continued)

TABLE 1 | Continued

References	Research designs of included studies	Number of participants	Participant characteristics	Controls	Outcome measures
Ng et al. (37)	3 prospective studies, 1 cross-sectional, pre-post case series	779	Not reported	General neurology clinic or general MND care	Survival, hospital readmissions and length of stay, SF-36, VAS on life satisfaction and well-being, ALSSS, ALSFRS, CSI, healthcare costs
Dal Bello-Haas and Florence (4)	2 studies, 6 and 12 month parallel group (1 randomized and 1 quasi randomized trial)	52	27 people with definite or probable, probable with laboratory-supported MND (El Escorial criteria), aged 41–80 years. Early stage MND. 25 people with definite or probable MND (El Escorial criteria), aged 41–80 years. Mild to moderate stages of MND.	The control condition was either no exercise or standard rehabilitation management (for example, range of motion exercise or stretching exercise).	LSFRS/the SF-36 to measure quality of life/FSS/Manual muscle strength testing
Eidenberger and Nowotny (34)	2 RCTs, 1 pre-experimental study and 1 with a historical control group	87	Male, *N* = 57; Females, *N* = 30 Mean Age (Y): range 53–63 years	Sham training/historical controls/no controls/ lowest possible load	Respiratory-related OMs (e.g., Spirom/FVC/MIP/MEP etc.)/Total survival time/6 MWT/Hand-held dynamometry/ ALSFRS/ FSS/HRSD/ESS/FIM/EQ-5D/SF-36/Chronic Respiratory Questionnaire
Arbesman and Sheard (36)	2 RCTs/2 non-RCTs + single subject study	287 (including Dal Bello-Haas and Florence (4))	Only one study (i.e., single-case study) reported: one male, age 62 years. Drory et al. (45) and Bello-Haas et al. (46) and already presented in previous systematic reviews.	Training vs no training/or general care	ALSFR/ Medical Outcomes Survey 36-item/QoL: SF−36/Life satisfaction and well-being/visual analogue scales ROM/Muscle strength and shortness/Grip strength/Functional activities—Modified Norris Limb Scale/Muscle strength measured with Chatillon push–pull gauge/Survival
Ferreira et al. (35)	3 RCTs	63	Not reported	Comparison with controls who had not received RMT full time or were receiving training without load	Ventilatory function FVC, FEV1, MVV/Respiratory muscle strength, MEP and MIP)/Functional capacity, 6 MWT
Quinn and Busse (38)	4 studies with different designs: before/after design (*N* = 1), single case (*N* =1), observational (*N* = 1), RCT (*N* =1)	63	Male: female = 17:23 (only reported in 1 study - Zinzi et al. (48)). Age not reported except for one single case study = one male, 49 years old	Healthy controls/healthy matched controls/usual care (pharmacological)	Range of motion/Flexibility/Strength/Co-ordinated and reciprocal movement/Standing, one foot and kneeling balance/Breathing volume and control/SF-36/Number of falls/Modified falls scale/Berg Balance Scale/Self' paced/Fast paced gait speed/UHDRS/Physical examinations of posture/Zung depression scale/MMSE/Barthel Index (ADL)/Tinetti scale (balance)/PPT/Rehabilitation evaluation scale (REHAB)/BMD/Interact (behavior assessment)/HR/BP/RR/SHRS
Fritz et al. (39)	2 Observational (without control), 6 RCTs, 7 Pre-Post control group studies, 2 Pseudo RCT, 1 single case study	435	Male = 47.25% Mean Age (Y): range 28–57	Usual care (*N* = 2), no progression in resistance training (*N* = 1), sham (*N* = 1), otherwise not specified	Balance/Fitness (cardiovascular function)/Goal attainment/Motor function and performance/Muscle strength/Number of falls/Physical activity/Pulmonary function/Rate of chest infections/Ulcer staging/Spatiotemporal and kinematic parameters of gait and balance/Walking ability and endurance/Outcome measures of cognitive function included cognition and psychological measures (depression, anxiety, and apathy)
Koopman et al. (31)	3 RCTs (2 included in the scoping review)	120	One study was conducted in elderly people (no details provided). No details are reported in the other studies	No treatment or usual care	Self perceived activity limitations (e.g., Physical Component Summary of the SF-36 PCS/Physical mobility category of the Nottingham Health Profile)/Muscle strength/Muscle endurance fatigue/Pain/Adverse events subdivided into minor adverse events and serious adverse events
Trujillo-Martín et al. (40)	1 Clinical trial (pre-post design)	87	Mean age (Y), (SD) = 38.1 (10.9)	No controls	Neurological examination using the Romberg's Test and a coordination test with a computer

(Continued)

TABLE 1 | Continued

References	Research designs of included studies	Number of participants	Participant characteristics	Controls	Outcome measures
Fonteyn et al. (41)	14 prospective clinical trials (4 moderate quality i.e., comparative studies - 1 on cerebellar ataxia).	84	Not reported	Controls were patients receiving treatments later or not specified	Balance/gait/muscle strength/range of motion/ataxia severity/fall frequency/gait speed/ADL/FIM/Barthel Incapacitation scores/Hamilton Rating Scale for Depression/WHOQOL-BREF/NESSCA/SARA
Marquer et al. (43)	19 studies including MS and traumatic causes. In this scoping review only 4 were included: 1 RCT and 3 observational studies. However, 3 of these were already included in Fonteyn et al. (41). Only Foltz and Sinaki (49) is described in the scoping review.	19	Not reported	No controls	Subjective self-evaluation of balance
Milne et al. (42)	4 RCTs 1 Pseudo-RCT 4 Interrupted time series without a parallel control group 5 Case series	292 in total (21 were not adults) = 271	Mean age range (Y): 23.3–62.5 A total of 228 participants (out of 292) were ambulant, and 72 were non-ambulant. In 2 studies, ambulation status was not reported	No controls or pharmacological management alone or control group completing verbal health education and upper limb exercises (compared to a cycling regime) or a control group receiving sham vibration over the same duration (compared to stochastic vibration)	SARA/FIM/Gait speed/ cadence/ FAC/ Number of falls/ ICARS (8 items)/10 mWT/ Gait speed/ Standing capacity/ Spread of feet/ Body sway/ Knee to tibia test/ Action tremor/ SF-36/EQ-5D/EQ-VAS/ABC/SCAFI/INAS/GAS/BBS/Kinematic and kinetic gait parameters/Static balance test/ Dynamic balance acceleration treadmill task/DGI/ TUG/ FRT/ ABC/ Sway amplitude/ Spatiotemporal/ gait parameters/ FES-I LOS/ SOT/CoP area of 95% confidence/Ellipse CoP sway path/ CoP mean velocity/ Barthel WHOQOL-BREF/ MBI/ 5-item Barthel Index/ Obstacle avoidance task on a treadmill/ EFAP obstacle subtask/ Sway area
Hajjar and Cooper (44)	2 quasi randomized controlled trials	38 (two studies based on the same sample)	Not reported	Balance exercises only	Kinematic gait measures (stance time, swing time, and step length)/2.4-m walk test/Timed "Up & Go" Test/Vertical Gaze Fixation Score/Gaze Error Index

(Continued)

TABLE 1 | Continued

References	Research designs of included studies	Number of participants	Participant characteristics	Controls	Outcome measures
Intiso et al. (8)	6 case reports, 3 case series, one case-control study, one quasi randomized trial and one randomized controlled trial	88	Gender (number of F/M), information only in 3 studies: Case series 1: 6/2 Case series 2: 3/7 Case series 3: 2/3	Balance exercises only [same studies as in (44)]	BBS/ ABC Scale/Sharpened Romberg Test/ FRT/360 turns/ TUG test/ 6-MWT/10-WMT/15.2-meter walk test/8-foot (2.4-me) walk test/5-step test/Balance and gait parameters/ABF device/Static and dynamic baropodometry/Computerized systems including the GAITRite system/3D-GA/Force platforms/PSPRS/UPDRS

6 MWT, Six Minute Walking Test; RM, Repetition Maximum; VO2 max, Maximal Oxygen uptake; 6 MWD, Six Minute Walking Distance; ICU, Intensive Care Unit; CIS-fatigue, Checklist Individual Strength; MVC, Maximum Voluntary Contraction; MVIC, Maximum Voluntary Isokinetic Strength; PIPPS, Perceived Impact of Problem Profile Scale; ESS, Environmental Status Scale; CK, Serum creatine kinase; ALS, Amyotrophic Lateral Sclerosis; ALS-FRS, Amyotrophic Lateral Sclerosis Functional Rating Scale; EQ-VAS, EQ–Visual Analogue Scale; DGI, Dynamic Gait Index; LGMD1, Limb-girdle muscular dystrophies autosomal dominant; LGMD2, Limb-girdle muscular dystrophies autosomal recessive; MND, Motor Neurone Disease; RCT, Randomized Controlled Trial; PNF, Proprioceptive Neuromuscular Facilitation; SRV, Systematic Review; ALSFRS, Amyotrophic Lateral Sclerosis Functional Rating Scale; MT, Manual Muscle strength Testing; FSS, Fatigue Severity Scale; FIM, Functional Independence Measure; FVC, Forced Vital Capacity; BiPAP, Biphasic Positive Airway Pressure; MIP, Maximum Inspiratory Pressure; MEP, Maximal Expiratory Pressure; HRSD, Hamilton Rating Scale for Depression; ESS, Environmental Status Scale; EQ-5D, Health Status questionnaire; SF-36, Short Form-36 Health Survey; SNIP, Sniff Nasal Inspiratory Pressure; RMT, Respiratory Muscle Training; FEV1, Forced Expiratory Volume; MVV, Maximum Voluntary Ventilation; QoL, Quality of Life; CMTES, Charcot-Marie-Tooth; Examination Score; FITT, Frequency-Intensity-Time-Type; ROM, Range of motion; BMI, Body mass Index; FFM, Fat Free Mass; RMS, Root Mean Square; PCI, Physiological Cost Index; MHC, Myosin Heavy Chain; VAS, Visual Analogue Scale; METS, Metabolic Equivalent of Task; UHDRS, Unified Huntington's Disease Rating Scale; MMSE, Mini Mental Status Examination; PPT, Physical Performance Test; REHAB, Rehabilitation Evaluation Scale; BMD, Behavior and Mood Disturbance scale; HR, Heart Rate; BP, Blood pressure; RR, Respiratory Rate; SHRS, St Hans rating scale; ADL, Activity Daily Living; LOSWHOQOL-BREF, World Health Organization Quality of Life questionnaire; NESSCA, Examination Score for Spinocerebellar Ataxia; SARA score, Scale for Assessment and Rating of Ataxia; FAC, Functional Ambulation Classification; ICARS, International Cooperative Ataxia Rating Scale; ABC, Activities specific Balance Confidence; SCAFI, Spinocerebellar Functional Index; INAS, Inventory of Non-Ataxia Signs; GAS, Goal Attainment Score; BBS, Berg Balance Scale; TUG, Timed up-and-go test; FES, Falls Self-Efficacy Scale; LOS, Limits of stability; SOT, Sensory Organization Test; WHOQOL-BREF, WHO Quality of Life-BREF; EFAP, Emory Functional Ambulation Profile; RAM, Rotterdam Activity Monitor; FIS, Fatigue Impact Scale; HADS, Hospital Anxiety and Depression Scale; RHS, Rotterdam Handicap Scale; MMT, Manual muscle testing; DASS-21, Depression, Anxiety; Stress scale (short form); PIPP, Perceived Impact of Problem Profile; PEFR, Expected Peak Expiratory Flow Rate; FEV1, Forced Vital Capacity in 1 s; HRQoL, Health-related Quality of Life; MBI, Modified Barthel Index; MRS, Modified Rankin Scale; HDS, Hughes Disability Scale; NHP, Nottingham Health Profile; BI, Barthel Index; HAS, Handicap Assessment Scale; ALSSS, Amyotrophic Lateral Sclerosis Severity Scale; CSI, Caregiver Strain Index; BBS, Berg Balance Scale; ABC, Scale Activities-specific Balance Confidence; FRT, Functional Reach Test; 10-WMT, Ten-Meter Walk Test; ABF, Audio-biofeedback device; 3D-GA, 3D-Gait Analysis; PSPRS, Progressive Supranuclear Palsy Rating Scale; UPDRS, Unified Parkinson's Disease Rating Scale; FSHD, Facioscapulohumeral Muscular Dystrophy; CoP, Centre of Pressure; OMs, Outcome Measures; FITT FDI, frequency, duration, and intensity score for the Phone-FITT scale.

TABLE 2 | Mapping to World Health Organisation international classification of function ICF) domains.

References	Condition included	Body-structure	Body-function	Activity	Participation	Disease-specific	Other
Voet et al. (23)	Muscle diseases (myotonic dystrophy, polymyositis and dermatomyositis, facioscapulohumeral muscular dystrophy, mitochondrial myopathy)		✓		✓		
Cup et al. (24)	Myotonic dystrophy and Welander distal myopathy		✓	✓	✓		
Gianola et al. (25)	Muscular dystrophy		✓	✓			
Narayanaswami et al. (26)	Welander distal myopathy, Becker muscular dystrophy, Limb-girdle muscular dystrophies, Hereditary inclusion body myopathies	✓	✓				
Habers and Takken (27)	Idiopathic inflammatory myopathy (dermatomyositis, polymyositis, and inclusion body myositis)	✓	✓		✓		
Ydemann et al. (28)	Critical illness myopathy and polyneuropathy		✓	✓	✓		✓
Young et al. (29)	Charcot-Marie-Tooth disease		✓	✓			
Sman et al. (5)	Charcot-Marie-Tooth disease	✓	✓	✓		✓	
Corrado et al. (30)	Charcot-Marie-Tooth disease	✓	✓	✓			
Quinlivan et al. (13)	McArdle disease	✓	✓				
Koopman et al. (31)	Postpolio syndrome		✓	✓			✓
Simatos Arsenault et al. (12)	Guillain-Barré Syndrome		✓	✓	✓	✓	
Khan and Amatya (32)	Guillain-Barré Syndrome		✓	✓	✓		
Dal Bello-Haas and Florence (4)	Motor-neuron disease		✓		✓	✓	
Lui and Byl (33)	Motor-neuron disease		✓			✓	
Eidenberger and Nowotny (34)	Motor-neuron disease		✓	✓	✓	✓	
Ferreira et al. (35)	Motor-neuron disease		✓	✓			
Arbesman and Sheard (36)	Motor-neuron disease		✓		✓	✓	
Ng et al. (37)	Motor-neuron disease				✓	✓	✓
Quinn and Busse (38)	Huntington's disease		✓	✓	✓	✓	
Fritz et al. (39)	Huntington's disease		✓	✓			✓
Trujillo-Martín et al. (40)	Spinocerebellar ataxia		✓				
Fonteyn et al. (41)	Cerebellar Ataxia		✓	✓	✓	✓	
Milne et al. (42)	Genetic degenerative ataxia		✓	✓	✓	✓	✓
Marquer et al. (43)	Cerebellar Ataxia		✓				
Hajjar and Cooper (44)	Progressive supranuclear palsy		✓	✓			
Intiso et al. (8)	Progressive supranuclear palsy		✓	✓		✓	✓

Step One and has been used in one RNC disease group and other neurological conditions (50). Three constructs (i.e., enjoyment, motivation and confidence) are important predictors of physical activity behavior and not assessed within any of the Ox-PAQ items but are relate highly to self-efficacy. The Sources of Self-Efficacy for Physical Activity (52) was thus identified as an additional secondary outcome able to reflect these constructs. Other self-efficacy scales found in the search were specific to particular diseases and populations but did not include neurological conditions. It is important to note that these outcomes were not only reflective of that which is important to stakeholders but also additionally are able to provide mechanistic insight for researchers.

Step Three: Quality and Feasibility Assessment of Proposed Outcome Measurement Instruments

The OxPAQ questionnaire is a short, 23-item, patient-reported outcome measure, that has been specifically developed for cross-disease application and validated in three long term neurological conditions (MND, Parkinson's disease, Multiple Sclerosis) (50). It was developed using patient interviews and expert reviews and has a manual and online scoring. The Ox-PAQ reports on three domains, Routine Activities (14 items), Emotional Well-Being (5 items) and Social Engagement (4 items). Routine Activities assesses individuals' capacity to engage in regular activities that form the basis of daily life. Emotional Well-Being provides an indication of current mental health status, while

Social Engagement assesses whether individuals can maintain relationships, both personal and from a wider community perspective. Internal reliability is high (Cronbach's α 0.81–0.96) and validity was demonstrated against relevant domains of the MOS SF-36 and the EQ-5D-5L (50). Sources of Self-Efficacy for Physical Activity is an 18-item questionnaire that measures six aspects (3 items for each source) of self-efficacy for physical activity, specifically: mastery experience, vicarious experience, verbal persuasion by others, self-persuasion, negative affective states and positive affective states (52). Items were pooled from prior qualitative studies, scales of feelings induced by physical activity and sources of self-efficacy more generally. It was refined in a study of 1,406 German adults through principal axis analysis with inter-related factors and confirmatory factor analysis. It is a reliable (Cronbach's α 0.75–0.93), valid (convergent and discriminant).It has not been validated in neurological populations, but the scale was designed to be generally inclusive allowing it to be applied across conditions and populations (52). Other self-efficacy scales target specific conditions and were not generalizable or applicable to people with RNC.

Step Four: Reaching Consensus on the Proposed Core Outcome Set

Following broad group communication and discussions and a final face to face consensus procedure involving small group discussions, it was agreed that the Ox-PAQ and the Sources of Self-Efficacy for Physical Activity measure should be assessed in trials evaluating physical activity interventions across RNCs given the ways in which they matched the domains and constructs of importance identified by the stakeholder group. This was a group decision by people living with RNC, charity representatives and the research team at the final workshop.

DISCUSSION

Physical activity research trials in RNCs to date have typically involved targeted exercise intervention and evaluation at the specific disease level despite these diseases leading to variable but similar impairments and functional impacts (for example fatigue, muscle weakness, balance problems, falls and difficulty walking). Our scoping review highlighted the prevalence of interventions, mainly focusing on structured exercise and typically underpinned by standard approaches (53) highlighting the role of physical activity and exercise as a critical enabler of participation for all those living with common and rarer long term neurological diseases (54). Our scoping review of the literature identified outcome measures appropriate for the specific body structure, function and activity level changes targeted by these interventions, but there was a degree of mismatch between these outcomes and constructs identified as important to people living with RNCs (e.g., assessments that capture changes at the level of participation).

We utilized a person-centered approach leading to the proposal of a meaningful core outcome measurement set for use when researching physical activity interventions for people living with RNCs. Our collaborative and participatory design involved members of the public, including people living with RNCs, representatives of charities and support groups for RNCs and is the first core outcome set to our knowledge which has specifically focused on physical activity interventions for RNCs. Stakeholder engagement is receiving increasing recognition in patient-reported outcomes research (18) and clinical trials (55, 56) so as to ensure that interventions and outcomes are relevant to the target populations. A core outcome set for disease modification trials for dementia has been developed with stakeholder input and involvement of the research community. This was achieved through a number of stages, including a systematic review of outcome measures, a consultation with patient and public involvement representatives and a final consensus reached with the dementia research community (20). A similar approach was used to develop a core outcome measure set for exercise studies in Multiple Sclerosis (57), where a group consisting of experts in the field, support group representatives and expert patients, jointly discussed a pre-defined core set for Multiple Sclerosis. This was based on the World Health Organisation International Classification of Function and included body structure and function, activity and participation categories. Our approach differed somewhat in that we initially elicited discussion and reflection from our stakeholder groups on the domains considered important when engaging in physical activity interventions, but without presenting any work undertaken in previous studies.

Outcomes identified in the scoping review assessed the effect of physical activity interventions primarily at the level of body functions and structures, functional activities. There were fewer identified outcomes at participation level, in contrast to the domains prioritized by our stakeholder group, namely physical and psychological well-being and participation to day-to-day activities. In the scoping review, measures of quality of life and health-related well-being were identified, but these did not (in the views of our stakeholder group) sufficiently capture the breadth of areas of importance in relation to participation and physical activity in RNCs. For example, the 36-Item Short Form Survey is more focused on levels of vigorous and moderate activities, rather than independence in day-to-day activities. The Ox-PAQ and the Sources of Self-Efficacy for Physical Activity measure were however considered to reflect meaningful outcomes of physical activity interventions for people with RNCs.

Whilst the identified and proposed outcomes are clearly relevant to people with RNC, it is not yet clear how well the measures perform within and between these populations nor whether they fully capture that which is meaningful to people with RNCs. For example, the Sources of Self-Efficacy Scale may not fully capture enjoyment for physical activity; it may be that a purpose developed enjoyment scale (58, 59) is more appropriate in different settings. The broad range of rare neurological diseases where physical activity interventions are indicated are a specific challenge. A key limitation is that we did not consistently have stakeholders present at all workshops with faster progressing conditions, those with significant cognitive disorders or carers, relying on the charity representatives to bring accounts of these experiences. People were invited, but the additional complexity of those conditions may have affected engagement in all steps. Future validation work will need to include these groups to inform the implementation of the proposed core outcome set.

CONCLUSION

We propose a core outcome set, developed in collaboration with people living with RNC and their representatives, for use in studies of physical activity interventions. The two measures proposed were selected to include domains of importance to people living with these diseases.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/supplementary material, further inquiries can be directed to the corresponding author/s.

ETHICS STATEMENT

Ethical review and approval was not required for the study on human participants, in accordance with the local legislation and institutional requirements.

AUTHOR CONTRIBUTIONS

GR and MB: conception and organization of the research project, design and review and critique of the analysis, and writing of the first draft and review and critique of the manuscript. VB: organization of the research project, design and review and critique of the analysis, and writing of the first draft and review and critique of the manuscript. HD: conception and organization of the research project, design of the analysis, and writing of the first draft and review and critique of the manuscript. AB, TJ, JM, LP, RP, MR, and LR: conception of the research project, design of the analysis, and review and critique of the manuscript. FJ and ER: conception and organization of the research project, design of the analysis, and review and critique of the manuscript. All authors contributed to the article and approved the submitted version.

FUNDING

This work was funded by an NIHR Programme Development Grant RP-DG-0517-10002 (Co-Chief Investigators: GR and MB). This is a summary of independent research funded by the National Institute for Health Research (NIHR)'s Programme Development Grant Programme. Centre for Trials Research receives funding from Health and Care Research Wales and Cancer Research UK. Open Access funding has been made available from Cardiff University.

ACKNOWLEDGMENTS

We would like the acknowledge the input of our stakeholder groups and charity representatives from the Muscular Dystrophy UK, Ataxia UK, Hereditary Spastic Paraparesis support group, Progressive Supranuclear Palsy Association, Huntington's Disease Association of England and Wales, Multiple Systems Atrophy Trust, Motor Neuron Disease Association. The scoping review that formed part of this work was registered on the Open Science Framework https://osf.io/4cr32/.

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Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers
in Rehabilitation Sciences

ORIGINAL RESEARCH
published: 15 November 2021
doi: 10.3389/fresc.2021.707612

Using a Co-design Approach to Create Tools to Facilitate Physical Activity in Children With Physical Disabilities

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Specialty section:
This article was submitted to Disability, Rehabilitation, and Inclusion, a section of the journal Frontiers in Rehabilitation Sciences

Received: *10 May 2021*
Accepted: *18 October 2021*
Published: *15 November 2021*

Citation:
Bolster EAM, Gessel Cv, Welten M, Hermsen S, Lugt Rvd, Kotte E, Essen Av and Bloemen MAT (2021) Using a Co-design Approach to Create Tools to Facilitate Physical Activity in Children With Physical Disabilities. Front. Rehabilit. Sci. 2:707612. doi: 10.3389/fresc.2021.707612

Introduction: There is a lack of effective interventions available for Pediatric Physical Therapists (PPTs) to promote a physically active lifestyle in children with physical disabilities. Participatory design methods (co-design) may be helpful in generating insights and developing intervention prototypes for facilitating a physically active lifestyle in children with physical disabilities (6–12 years).

Materials and methods: A multidisciplinary development team of designers, developers, and researchers engaged in a co-design process–together with parents, PPTs, and other relevant stakeholders (such as the Dutch Association of PPTs and care sports connectors). In this design process, the team developed prototypes for interventions during three co-creation sessions, four one-week design sprint, living-lab testing and two triangulation sessions. All available co-design data was structured and analyzed by three researchers independently resulting in themes for facilitating physical activity.

Results: The data rendered two specific outcomes, (1) knowledge cards containing the insights collected during the co-design process, and (2) eleven intervention prototypes. Based on the generated insights, the following factors seem important when facilitating a physically active lifestyle: a) stimulating self-efficacy; b) stimulating autonomy; c) focusing on possibilities; d) focusing on the needs of the individual child; e) collaborating with stakeholders; f) connecting with a child's environment; and g) meaningful goal setting.

Conclusion: This study shows how a co-design process can be successfully applied to generate insights and develop interventions in pediatric rehabilitation. The designed prototypes facilitate the incorporation of behavioral change techniques into pediatric rehabilitation and offer new opportunities to facilitate a physically active lifestyle in children with physical disabilities by PPTs. While promising, further studies should examine the feasibility and effectivity of these prototypes.

Keywords: tools, qualitative data, physical disabilities, children, co-design, physical activity

BACKGROUND

The benefits of encouraging a physically active lifestyle from an early age have been consistently documented (1). A meta-analysis showed an association between higher levels of physical activity and lower morbidity for typically developing children (2), and this is also assumed for children with physical disabilities. Moreover, physical activity is positively associated with psychosocial health for children with physical disabilities, including self-perceptions and health-related quality of life (3–5). However, most children, especially those with physical disabilities, do not meet the Dutch physical activity guidelines of at least 60 min of moderate-intensity activities every day (6–8).

While pediatric physical therapists (PPTs) are the designated professionals to facilitate physical activities in children with disabilities (9), a systematic review showed the lack of effective interventions available for PPTs to increase physical activity levels in these children (10). Increasing physical activity in children with disabilities is complex and multi-faceted, because of the variety of personal and environmental factors influencing physical activity (11–13). Important personal barriers are a lack of self-confidence, and feeling like an outsider (11). An important environmental barrier is the inability of people to see possibilities for children with disabilities to become physically active (11). Specific behavioral change strategies might provide PPTs with the right tools to support children and their parents to overcome personal and environmental barriers that hinder them to participate in physical activities (12, 14).

Appropriate opportunities at sports clubs and in the general community are important environmental requirements for children with disabilities to achieve a physically active lifestyle (11, 13). In the Netherlands, care sports connectors (CSC) aim to create opportunities for children with and without physical disabilities to become physically active, and in addition connect physical activity and sports professionals with health care sectors (15). Unfortunately, as yet, the collaboration between PPTs and CSCs has been lacking (16); sports participation in children might increase when collaborations between these two professions improve.

A disturbingly large part of interventions developed during scientific research are not used in clinical practice (17). This may be explained by a lack of attention for stakeholder acceptability and implementation in existing practices during the development of healthcare interventions (18). Actively engaging stakeholders, such as children, parents and healthcare providers, throughout all stages of intervention development could increase the actual use of healthcare interventions (19). Co-design, defined as collective creativity across the entire design process (20), is a design thinking methodology that has the potential to lead to the development of interventions that are more engaging, satisfactory, and useful to potential end-users. During co-design, an active collaboration occurs between researchers, designers, developers and end-users "as experts of their own experiences" (21), and, done rightly, co-design brings together their different views, input and competences (22). Knowledge transfer between stakeholders is important when developing and designing new interventions and co-design is characterized by incremental knowledge over time during a project (23, 24). Based on existing knowledge and generated insights, stakeholders can create principles for interventions. These principles can be transitioned into ideas and furthermore in testable prototypes. Because of its potential for improving implementation of newly developed interventions, co-design should be further examined in healthcare. Therefore, at first, examples of good practices are essential to increase knowledge about how co-design can be successfully applied in the development of interventions in pediatric rehabilitation (10).

This study presents a co-design approach for the development of prototypes containing behavioral change strategies for PPTs to facilitate physical activity in children with disabilities and a prototype to optimize collaboration between PPTs and CSCs. The first aim of this study is to describe the insights generated during co-design related to "facilitating physical activity." The second aim is to describe the prototypes designed during co-design, based on knowledge from evidence and the generated insights during this method, to facilitate physical activity in everyday life settings of children with physical disabilities (6–12 years).

METHOD

Design

The present case study, called "What moves you?!" (funded by a grant from SIA, the Netherlands Taskforce for Applied Research, number RAAK.MKB08.006.), used different co-design methods to generated insights, and design and develop intervention prototypes. Collective decision-making and knowledge transfer between participating stakeholders was important during this process. Therefore, the principles of participatory action approach (PAR) were followed in this study. In PAR, stakeholder inclusion is extremely important and realized through active collaboration between stakeholders and researchers and there is a transfer of knowledge over multiple iterative development cycles (25), which is in accordance with co-design methods (26). Roughly, the co-design methods contained (1) three co-creation sessions, (2) four one-week design sprints (27–29) (3) living lab testing after each design sprint and (4) two triangulation sessions (**Figure 1**).

Participants

Our consortium ($n = 51$) consisted of a broad range of stakeholders, such as parents of children with physical disabilities ($n = 4$), adults with a physical disability ($n = 2$), PPTs ($n = 18$), CSCs ($n = 8$), and others such as members from the Dutch parent association for children with a disability ($n = 1$), the Dutch Association of PPTs ($n = 1$), Fitkids Foundation ($n = 2$) which has the responsibility to ensure the quality of an exercise therapy program for children with a chronic condition or disability in the Netherlands; (30), the Knowledge Centre for Sports & Physical Activity Netherlands ($n = 1$), Special Heroes ($n = 1$) (Dutch organization promoting a healthy and active lifestyle for people with a disability), designers/developers ($n = 2$) and researchers ($n = 12$). A core team was responsible for planning, preparation, and execution of the co-design process including co-creation sessions, the four one-week design sprints, and the triangulation sessions. The core team of this project consisted of researchers

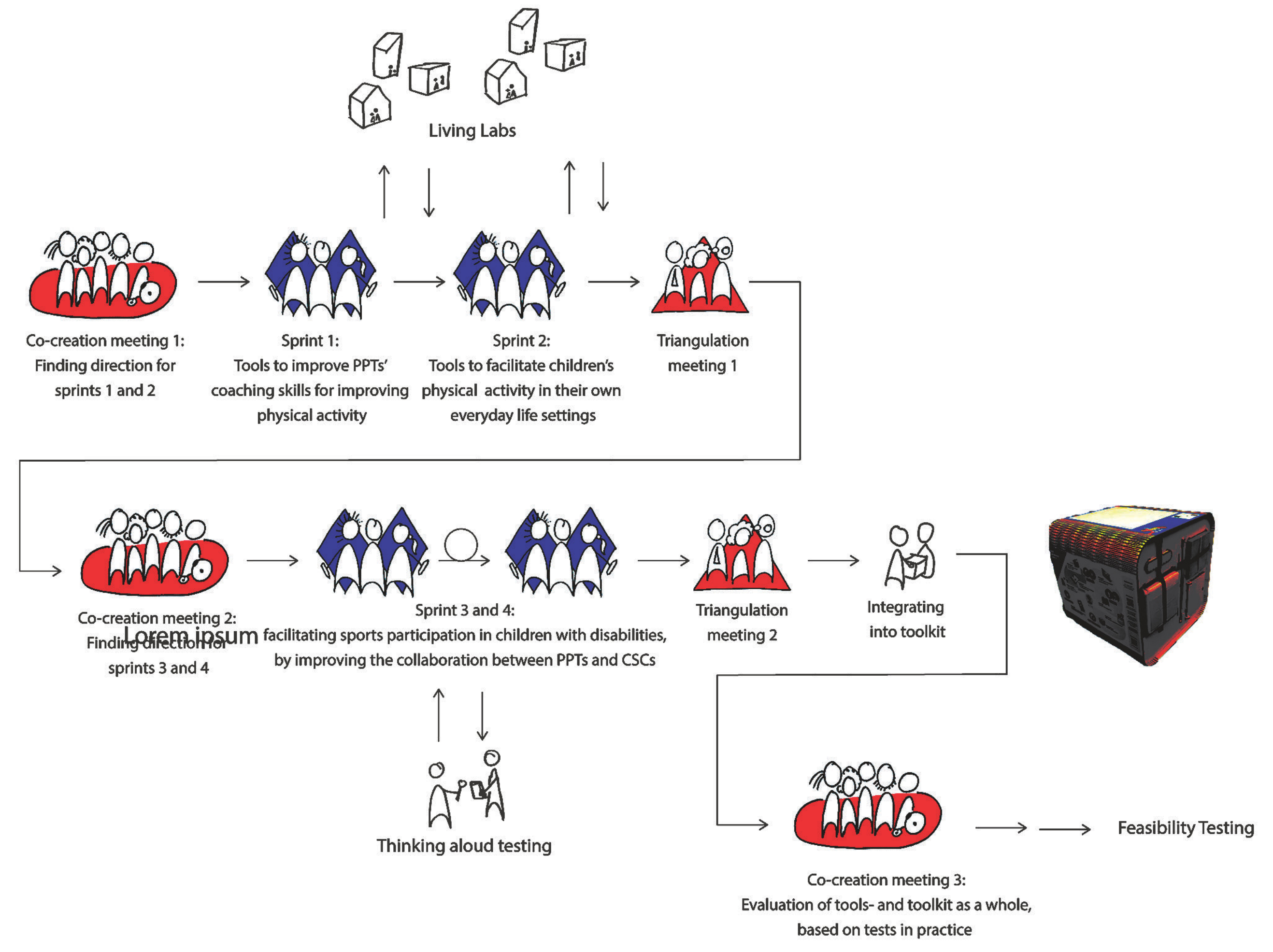


FIGURE 1 | A visual representation of the co-design process containing three co-creation sessions, four one-week design sprints, living lab testing, and two triangulation sessions.

with a background in PPT ($n = 2$), behavioral science ($n = 2$), design ($n = 2$), and social work ($n = 1$) and designers ($n = 1$). The consortium engaged in the co-creation sessions, furthermore PPTs and CSCs from our consortium tested the first prototypes of the tools in their own practice (living lab). Ethical approval for this study was granted by the healthcare ethics committee of the University of Applied Sciences Utrecht (99_000_2019).

Co-creation Sessions

During the preparatory phase, the core team ($n = 8$) collected insights from literature and practice. The core team demonstrated these insights during co-creation sessions and evaluated if these insights resonated with the different stakeholders from the consortium, and if they could relate to these insights with their own (professional) experience. Furthermore, the core team used different generative techniques, such as mapping sessions, during co-creation sessions to evoke tacit knowledge and latent needs (21). *Mapping* is a method to create a visual representation of interacting variables to facilitate the understanding of complex systems (21). These methods were used to explore the ideas, needs and values from stakeholders beyond their first response by collecting, for example, their wishes, dreams and barriers for potential interventions. By using generative methods, the core team empowered a large variety of stakeholders to participate during co-creation sessions; this stimulated and improved knowledge transfer, with an increase of insights over time.

After the first co-creation session, the core team defined the focus for the sprints based on knowledge from evidence and insights generated during this session. During the first sprint, the team focused on the development of prototypes to improve PPT's coaching skills for improving physical activity. During the second sprint, the team focused on the development of prototypes to facilitate children's physical activity in their own everyday life settings. During the third and fourth sprint, the core team focused on facilitating sports participation in children with disabilities, by improving the collaboration between PPTs and CSCs.

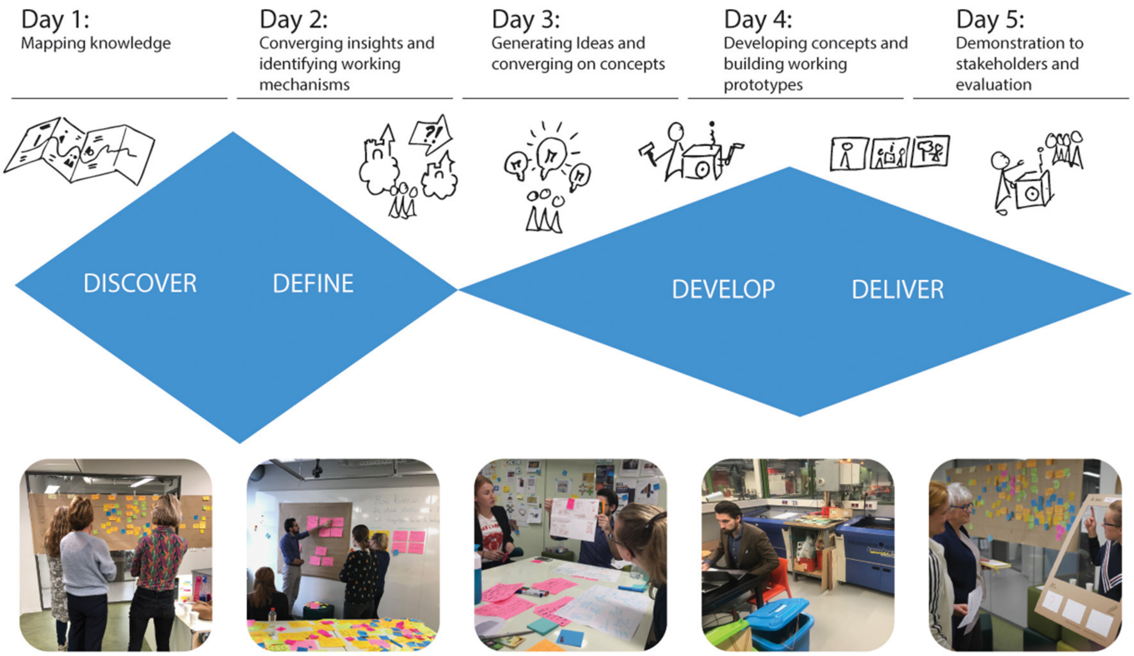


FIGURE 2 | A visual representation of a 5 day sprint week.

Sprint Weeks

To design the prototypes, the core team ($n = 8$) set up four design sprint weeks based on the Google Design Sprint approach (28). This approach consists of a 5 day process for answering critical development questions through design, prototyping, and testing ideas with stakeholders. The goal of each sprint was to quickly develop feasible prototypes for interventions based on knowledge from evidence, generated insights during this project and user testing, with maximum attention to stakeholder participation. We used the adopted Double Diamand described by Elbers et al. (19) for the sprint weeks containing two sequences of diverging and converging. See for a visual representation of the sprint week (**Figure 2**).

On the first sprint day, the core team went through a divergent phase in which they collected and mapped available knowledge from literature, practice and co-creation sessions with different co-design methods such as context mapping sessions, user journeys and socionas. The *user journeys* enable stakeholders to collaboratively construct a timeline that illustrates the journey of a child with physical disabilities and a goal related to increase physical activity from the start of PPT treatment (31). *Socionas* are a tool to stimulate designers to incorporate the systemic view around a child into the design process (32). A sociona consists of a visual representation of the dynamics in a system of people (for example children, parents, PPTs, CSCs on micro level and stakeholders setups on macro level) (32). After collating the insights, the core team performed further user research on day one with stakeholders (involving 4–8 stakeholders depending on the sprint week) from the consortium for instance by performing in-depth interviews with stakeholders. Stakeholders also had the opportunity to reflect on the collected data.

The second sprint day focused on converging activities, by selecting emergent themes from the insights gathered on day one. Based on these themes we determined the main working mechanism driving the behavior change intended by the interventions. For example one of the targeted working mechanisms was "support children in creating their own solutions." The other targeted working mechanisms are described in **Figure 2**. Determining such working mechanisms is important in prototype development, because they give insights in the expected efficacy of the intervention.

On the third sprint day, using different brainstorming techniques, the core team went through a divergent phase, by generating ideas for prototypes fitting these main working mechanisms. The team used the Behavorial Lenses Approach to integrate insights on individual determinants of behavior in the design activities (33). At the end of day 3 we focused on converging activities by selecting the concepts for the most promising prototypes using guiding principles for the interventions developed on the first sprint day.

On day 4, the designers of the core team ($n = 3$) developed working versions of the prototypes to make the working principles tangible (34). Each sprint finished on day 5, with a demonstration lunch in which the core team presented the prototypes to stakeholders from the consortium and colleagues. After the demonstration lunch, prototypes were adjusted based on their feedback. A reflective session, in which the core sprint team evaluated the sprint week, took place at the end of each sprint week.

TABLE 1 | Themes and subthemes.

Stimulating self-efficacy	Stimulating autonomy	Focusing on possibilities	Focusing on the needs of the individual child	Collaborating with stakeholders	Connecting with a child's environment	Meaningful goal setting
Fostering confidence	Being able to deny help	Focusing on abilities instead of obstacles	Using a tailored approach	Striving for equality	Have activities take place in daily life	Relevant goals
Fostering feeling secure	Knowing who is responsible	Creative solutions	Finding suitable solutions	Finding the right support	Have activities take place in a meaningful environment	Purposeful goals
Having insight in their own possibilities	Knowing their own boundaries	Having fun	Giving the child a central position	Sharing knowledge	Including the social environment	Goals focusing on participation
Being motivated	Being able to create their own solution	Challenging solutions	Listening to each other	Monitoring the child	Fostering visibility	
	Being able to try out activities	Small steps toward goal				
	Trial and error	Celebrating (actual) successes				

Living Lab Testing

At the end of sprint week 1 and 2, the prototypes were sent to fourteen PPTs to allow them to interact with the prototypes in their daily practice (34). They tested these prototypes together with children with physical disabilities and their parents. To reflect on these prototypes, structured telephone interviews with the PPTs were conducted after 4 weeks. During sprint three and four, the team designed (sprint 3) and developed (sprint 4) a mobile app to improve collaboration between PPTs and CSCs. This application was tested in a structured environment by children with physical disabilities, their parents, PPTs and CSCs to allow these participants to interact with this prototype. During these structured tests the participants were encouraged to "think aloud;" two or three observers documented this feedback.

Triangulation

To validate the prototypes, the core team organized triangulation sessions. Two behavioral scientists and two experts in social dynamic systems, all unrelated to the project, reviewed the prototypes, and especially their underlying working mechanisms. The behavioral scientists focused on the integration of behavioral insights in the prototypes. They identified which Behavioral Lens(es) they observed in the mechanism of the prototypes (33). The experts in social dynamic systems pointed out where prototypes responded to social aspects of behavioral change. All findings were then discussed by these experts and the core team.

Qualitative Analyzes

Three researchers with a background in PPT, design, and social work, independently structured and analyzed all available data from the "What moves you?!" study. The data consisted of sprint reports, reflective journals (daily self-reports in which the core team collected their experiences and thoughts on the co-design process) (35), and photos and film clips of co-creation sessions, sprint activities and triangulation sessions. Using Atlas.ti, we used a qualitative method to analyze our co-design data (36). An inductive thematic approach was used in which we coded fragments of text in step one, resulting in subthemes in step two (37, 38). In step 3, finally, we determined main themes. Step one consisted of defining a text or visual section as an important insight obtained during the co-design process. These insights should help answer three questions used to develop interventions during the four sprints (1) to improve PPT's physical activity coaching, (2) to facilitate children's physical activity in their own life settings, and (3) to improve collaboration between PPTs and CSCs in order to facilitate sports participation. Consensus between the three researchers was reached throughout this entire process. These themes, subthemes and quotes are also gathered in knowledge cards. Rather than solely disseminate knowledge among researchers through scientific articles we created these knowledge cards to ensure that the gathered insights from this study will reach and be used by PPTs.

After testing the prototypes in the living lab settings, the interviews conducted with PPTs ($n = 16$) were recorded and summarized. A content analysis was performed to determine barriers and facilitators in the usability of the tools. These barriers and facilitators were used to optimize the prototype of the tools.

RESULTS

At the end of this process there were two specific outcomes (1) generated insights collected during co-design, and (2) prototypes of the tools.

Insights

Table 1 presents an overview of the themes and subthemes gathered from all co-design data and the most important issues related to these themes and subthemes are discussed in the text below. The themes are included in the headings and both the themes and subthemes are in *italic* in the text below. The quotes represent the summarized translation of

FIGURE 3 | Example of knowledge cards with the theme *stimulating self-efficacy*.

what parents, adults with a physical disability, PPTs, CSCs, researchers and other stakeholders expressed during this co-design approach. Because of the intensive collaboration during this process it was not documented who was the author of the quote. **Figure 3** shows an example of a knowledge card with the theme *stimulating self-efficacy* and the subthemes *fostering confidence, fostering feeling secure* and *having insight in their own possibilities*.

Stimulating Self-Efficacy

For *stimulating self-efficacy*, *fostering confidence*, *fostering feeling secure*, *having insight in their own possibilities* and *being motivated* were pointed out as positive subthemes. Positive experiences for *fostering confidence* were described when children were able to move without assistance, for example when self-propelling their wheelchair instead of being pushed. *Fostering feeling secure* seems important when performing a sports, as mentioned by a CSC: "Feeling insecure when playing sports seems a reason for a child to walk away from a sports." *Having insight in their own possibilities* was pointed out as important when facilitating a physically active lifestyle: "We (parents, PPTs and CSCs) create feelings of insecurity and a delay in motor development if we can't achieve that children know what their own competences are."

Stimulating Autonomy

The importance of *stimulating autonomy* for children with a physical disability was mentioned often. Participants believed that, in order to become autonomous, it is important for children are able *to deny help*, *know their own boundaries* and *know who is responsible*. Where possible, children with a physical disability need the opportunity *to deny help*. However, parents and healthcare providers are often over-protective and over-supportive, and they often provide help immediately; as mentioned by a parent: "I find it hard to give my child the opportunity to deny help and become independent." Healthcare providers expressed the need to discuss *who is responsible* for achieving formulated therapy-goals: "it is important that I know where my responsibility stops and where the responsibility of a child, their parents or other healthcare providers starts." It is furthermore important for a child to *know what their boundaries are* when the goal is to become more physically active and autonomous: "by doing and discovering a child will experience their boundaries. So, dare to search for their real boundaries."

After becoming autonomous, the importance of staying autonomous was highlighted. A solution-oriented approach, where children are able *to create their own solutions*, seems a positive factor for being autonomous; as mentioned by a PPT: "I withdraw to see if the child comes up with his own solution," "I would rather act too late than too early to increase their exploratory behavior." This was also pointed out by an adult with a physical disability: "Often healthcare providers take control. For example, bus drivers often push children in a wheelchair to the school bus, even if these children are able to self-propel their wheelchair. What are the consequences of this behavior for a child's psyche?" The need *to try out new activities* was also mentioned: "By doing and discovering a child will discover their own limits." A solution-oriented approach is characterized by *trial and error*: "You should give children the opportunity to make mistakes, but without judgment if things go wrong."

Focusing on Possibilities

Focusing on *abilities instead of focusing on obstacles* is essential when stimulating a physically active lifestyle: "ask what a child can do instead of what they can't do." The way to achieve this is to create solutions in a child's own environment that are *creative*, *fun* and also *challenging*. The importance of *small steps* toward the final goal was highlighted in order to *celebrate (actual) successes*.

Focusing on the Needs of the Individual Child

When a (health) care professional is focused on facilitating a physically active lifestyle, it is important to *focus on the needs of the individual child* and their parents. The provided care must therefore have an *tailored approach* and the solutions for increasing physical activities must be *suitable* for a child and their environment: "Healthcare providers should sense what a child needs," "It involves customization, while protocols do not take the real needs of a child into account." The *child must have a central position* when providing care: "often we talk about children when we have to talk with children." This is only possible when stakeholders actually are *listening to each other* as mentioned by parents of a children with physical disabilities: "I wish professionals would really listen to parents in an open conversation without prejudice caused by the diagnosis," "It feels like fighting when I'm not heard."

Collaborating With Stakeholders

The importance of *collaborating with stakeholders* was commonly reported when discussing how adequate sports activities can be found for a child. Within the Netherlands CSCs and PPTs have the opportunity to collaborate when searching for sports, together with children and their parents. During this collaboration it is important that conditions are created in which all stakeholders *feel equal*. *Sharing knowledge* is one of the key ingredients to strive for equality: "The PPT probably knows better what the possibilities of a child are, but the CSC often knows more about relevant sports activities." *Finding the right support* for a task is often challenging for children and their parents and also for healthcare providers: "It is difficult to find the right healthcare provider who can guide the child toward a sports." As mentioned before, children with a disability often walk away from sports. Therefore, *monitoring the child* when starting and playing sports is important "dropping out from a sports might also be good, a child has tried and we now know that this does not work."

Connecting With a Child's Environment

Connecting with a child's environment was often mentioned when discussing how to facilitate children's physical activity in their own life settings. First of all, interventions should focus on being active in *daily life* situations; as explained by a PPT: "You try to provoke the child to move differently in their own environment." Therefore, PPTs must leave their own practice and include the *meaningful environment* of a child in their routine: "A success factor is going outside, into a child's own environment," "during the treatment" and "at home" are two different worlds." *Including the social environment* is another key ingredient when facilitating a physically active lifestyle: "Involving parents is not just letting parents watch, but let them participate and experience," "Friends of a child sometimes come to my treatment so that these children can learn skills together and integrate this activity at home (for example when playing tag)." If a child wants to *connect with their environment* it is important that a child is *visible*, so that, for example, other children in their own environment know who they are. This was explained by an adult with a physical disability: "It is important to make yourself visible to other children in your own environment. The older you get, the more difficult this is."

Meaningful Goal Setting

Meaningful goal setting was often mentioned as one of the most important aspects of a healthcare intervention. For children, parents and their healthcare providers it is important that goals are *relevant* and *purposeful* and that the main goal of the therapy is focused on *facilitating participation*. When goals are *relevant* this will motivate children and their parents to achieve their goals: "it is important to set goals for the intervention together with children and their parents." Goals should be *purposeful*: "it is important that goals are clear for children and their parents and not vague." Furthermore, because increased participation in physical activities should be the main focus of an intervention, goals should ideally be set on "participation" level; "the main goal should focus on participation," "during an intervention, don't solely focus on activities such as walking, but focus on participation, for example moving from one place to another."

Prototypes of the Tools

Eleven tool prototypes were designed during the sprint weeks (**Table 2**): four physical tools to improve PPT's physical activity coaching, four physical tools and two information videos to facilitate children's physical activity in their own life settings, and a mobile app to improve collaboration between PPTs and CSCs.

DISCUSSION

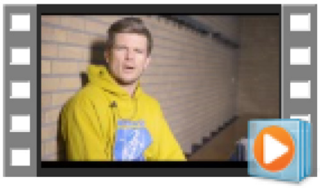
This study shows how a co-design approach can be successfully applied to generate insights and develop interventions in pediatric rehabilitation. The study had two aims: firstly, to describe all insights on how PPTs, CSCs, parents, and others can support children with physical disabilities in active lifestyles, obtained during the co-design process. The second

TABLE 2 | The designed tools including pictures, the targeted working mechanism and a description of the tools.

Prototype	Picture	Targeted working mechanism	Description
My Diary to improve PPTs' physical activity coaching		Integrate the social and meaningful everyday context of children in a PPT's treatment.	A diary for a child and their parents (separately) to track the amount of help a child is getting / parents are giving during a single day. This diary can be discussed during a PPT's session with a child and their parents to create awareness of the existing habits.
Look through the Window to improve PPTs' physical activity coaching		Facilitate a positive, observing role for parents during a PPTs treatment.	Parents can hold this window during a PPTs sessions. Instead of interfering in a conversation and/or intervention, they are invited to observe their child and discuss their findings afterwards. All questions encrypted in the window are positively formulated.
Question Dice to improve PPTs' physical activity coaching		Support children in creating their own solutions.	These question dice help a child create and try their one solution. After rolling the dice, the child is confronted with a question that stimulates a creative solution, e.g., "how would your superhero achieve this?"
Fears, Dreams, Actions Card set to improve PPT's physical activity coaching		Support parents in releasing their child.	This card set helps to discuss the fears and obstacles that a child and/or their parents might have when setting a meaningful goal. After discussing their fears, a child's and parent's dreams are discussed. Based on these dreams, the PPT, parent and child can formulate actions to achieve their goals.
Conversation placemat to facilitate children's physical activity in their own life settings		Provide insight into a child's opportunities and obstacles in their own environment.	This placemat helps to create a better understanding of the social and physical environment of a child. 3D figures (persons, houses, trees, cars, wheelchairs, etc.), can be placed on the placemat and a child can write or draw on the placemat. The child, their parents and the PPT can create a visual overview of the child's environment. Together with child and parents, the PPT can discuss opportunities and obstacles in a child's own environment.

(Continued)

TABLE 2 | Continued

Prototype	Picture	Targeted working mechanism	Description
Key ring to facilitate children's physical activity in their own life settings		Help a child refuse unwanted aid actively.	A child can attach this key ring to their clothes, backpack, or any other spot. Different labels are attached with messages such as "look at me, I did it myself!" and "you won't help me by helping without asking". The child can pull of a part of the label and present this to the person who wanted to help without asking.
Stickers to facilitate children's physical activity in their own life settings		Help a child refuse unwanted aid passively.	The stickers have messages like "yes, ask me what I need," and "I'm my own superhero". The stickers can be placed on a wheelchair, backpack, clothes etc. The stickers have a creative design and the messages are positively formulated.
Clapboard to facilitate children's physical activity in their own life settings		Improve the clinical handover between healthcare providers with a specific role for a child and his parents.	The video frame gives a child and his parents the opportunity to present their own goals of a healthcare intervention to healthcare providers with a video. Or to show, with a video, what a child is capable of. This improves the handover between e.g., PPTs and doctors, or PPTs and teachers.
Information video's to facilitate children's physical activity in their own life settings		Inform children, parents and healthcare providers about the positive effects of an active lifestyle.	The videos discuss the effects of stimulating self-efficacy by, for example, refusing unwanted help and the importance of connecting with the environment. Both videos are created by adults with a physical disability.
Application what moves us? to improve collaboration between PPTs and CSC		Improve collaboration between PPTs and CSCs in order to facilitate sports participation for children.	The application makes it possible for PPTs and CSCs to search for PPTs and/or CSCs in their community (through a google map overlay). They can link a sports professional to a child, and they can track the progress of a child when searching and performing a sports.

aim was to describe the interventions designed in the co-design process. Regarding the first aim we found the following positive factors of importance for children, their parents and (health)care professionals: (1) stimulating self-efficacy, (2) stimulating autonomy, (3) focusing on possibilities, (4) focusing on the needs of the individual child, (5) collaborating with stakeholders, (6) connecting with a child's own environment, and (7) meaningful goal setting. Regarding the second aim, to describe the designed intervention prototypes based on evidence and generated insights from the co-design process: these designed prototypes focus on determinants of behavior (i.e., self-efficacy and autonomy), possibilities, and connecting with a child's own environment. These intervention offer new opportunities to PPTs and CSCs to support children with physical disabilities in obtaining a more physically active lifestyle.

The generated insights during this co-design process reflect the importance of creating interventions aiming at behavior for facilitating physical activities in children with physical disabilities; as yet, hardly any such interventions exist (10). The shift from improving functions (i.e., physical fitness) and activities (i.e., motor skills) toward supporting determinants of behavior by PPTs was also underlined in a recent study of Reedman et al. (14). When facilitating a physically active lifestyle, an individually tailored approach, focusing on the needs of the individual child (11, 14), and setting meaningful goals (14, 39–41) are often mentioned in the literature (as well as in this study), however, the shift toward behavioral support is rather new in pediatric rehabilitation. Reedman et al. concluded that clinicians should, for example, focus on optimizing motivation and stimulating self-efficacy (14). Stimulating self-efficacy, by increasing confidence, security, and motivation, was also mentioned by many of our stakeholders–parents, adults with physical disabilities and PPTs. Furthermore, stimulating autonomy was often mentioned during our co-design activities. To become and stay autonomous, it is important that a child gets to know their own boundaries, that they are able to deny help, and that they can create their own solutions rather than adopting the solutions provided by parents or healthcare professionals. In sum, (health) care providers, such as PPTs, should focus on supporting determinants of health behavior when facilitating a physically active lifestyle.

One of the main competences of a PPT is creating fun and playful interventions for children. Having fun while being active in daily life activities and sports is very important for increasing leisure-time activities. As a consequence, having fun might increase physical activity levels (42). Stakeholders in this study underlined that focusing on abilities rather than obstacles is important for a PPTs intervention and that having fun, being creative and celebrating (actual) successes should be integrated in their interventions. However, PPTs should make a shift from creating a fun environment in a PPT's session to creating fun in everyday physical activities (43). Connecting with the everyday environment, and integrating the meaningful and social environment in their interventions, was mentioned as important and difficult. Darrah et al. (44) created a context approach were therapists are trained in changing tasks and environmental factors rather than changing the abilities of a child. When using this context approach it is important that the interventions take place in the natural environment of a child, while PPTs interventions are still mostly taking place in their own practices.

Stakeholders mentioned the importance of supporting behavior, focusing on possibilities and providing therapy in the natural environment of a child is important, as confirmed by literature (43). However, PPTs in this project also mentioned a lack of knowledge and tools to focus on these elements during their interventions. Furthermore, literature shows that logistics, time and (as a consequence) costs make it difficult to provide therapy in the everyday environment of a child (44). Rather than solely disseminate knowledge among researchers through scientific articles we created knowledge cards to ensure that the gathered insights from this study will reach and be used by PPTs. Furthermore, we designed and developed tools focusing on behavior and connecting with the environment. While the generated insights during this co-design approach provides directions for a PPTs intervention, we have not yet evaluated the efficacy of the designed tools. Therefore, the next step is to combine these tools in one toolbox and conduct a feasibility study and then an effectiveness study, to examine whether this toolbox actually increases PPTs efficacy to facilitate physical activity in children with physical disabilities.

One of the strengths of a co-design approach is the possibility to include many stakeholders with different backgrounds, as done in this study. However, co-design is time consuming (45), and capturing and documenting the knowledge transfer during co-design is difficult because the amount of data and the different sorts of data (e.g., interviews, photos of mapping sessions). During this project one researcher was responsible for collected all available data and therefore the knowledge was captured and documented carefully and, while structuring and analyzing the data was time consuming, the generated insights during this project provide a wide overview of expert knowledge related to the theme "facilitating physical activity." However, because of the active collaboration between different stakeholders the author of a quote is not documented. Therefore, the data does not represent separate views from parents, adults with a physical disability and professionals.

CONCLUSION

A co-design approach is an effective way to generate insights and explore new interventions for healthcare providers such as PPTs and CSCs. They can benefit from this co-design approach because it affords a better understanding of their needs. The designed prototypes facilitate the incorporation of behavioral change techniques into pediatric rehabilitation and thereby offer new opportunities to facilitate a physically active lifestyle in children with physical disabilities. Our findings suggest that when facilitating a physically active lifestyle, it is important to focus on (1) stimulating self-efficacy, (2) stimulating autonomy, (3) focusing on possibilities, (4) focusing on the needs of the individual child, (5) collaborating with stakeholders, (6) connecting with a child's own environment, and (7) meaningful goal setting.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

Written informed consent was obtained from the consortium partners for the publication of any potentially identifiable images or data included in this article.

AUTHOR CONTRIBUTIONS

EB, CG, MW, SH, RL, EK, AE, and MB were all responsible for data collection. EB, CG, and MW performed the qualitative data analyses with MB as supervisor. EB and MB were primarily responsible for writing the article but all other authors contributed also. SH, RL, and MB contributed to the study design.

FUNDING

Parts of this research were funded by SIA, the Netherlands Taskforce for Applied Research No. RAAK.MKB08.006. One Planet research Center was funded by the Province of Gelderland.

ACKNOWLEDGMENTS

We thank all children, parents, pediatric physical therapist and care sport connectors for their feedback on our tools. We also thank Vera Langeveld, Nienke Pruntels, Marloes Hagel and Margreet Sanders for their assistance in the development phase during their graduation projects.

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Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers
in Rehabilitation Sciences

ORIGINAL RESEARCH
published: 10 December 2021
doi: 10.3389/fresc.2021.726436

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Specialty section:
This article was submitted to Disability, Rehabilitation, and Inclusion, a section of the journal Frontiers in Rehabilitation Sciences

Received: *16 June 2021*
Accepted: *02 November 2021*
Published: *10 December 2021*

Citation:
Fortune J, Norris M, Stennett A, Kilbride C, Lavelle G, Hendrie W, DeSouza L, Victor C and Ryan JM (2021) Correlates of Objectively Measured Physical Activity Among People With Multiple Sclerosis: A Cross-Sectional Study. Front. Rehabilit. Sci. 2:726436. doi: 10.3389/fresc.2021.726436

Correlates of Objectively Measured Physical Activity Among People With Multiple Sclerosis: A Cross-Sectional Study

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Background: Identifying correlates of physical activity (PA) for people with multiple sclerosis (MS) is essential to design effective PA interventions.

Methods: Participants completed a battery of questionnaires and wore an ActiGraph accelerometer. Light physical activity (LPA) and moderate-to-vigorous physical activity (MVPA) (min/day) were calculated. Associations were examined using multiple linear regression adjusted for demographic and clinical confounders.

Results: Fifty-eight adults with MS participated (mean ± SD age: 56.8 ± 9.2 yr; 67% women). MS type was associated with time in LPA. Participants with secondary progressive MS ($B = -54.0$, 95% CI −84.7 to −23.3) and primary progressive MS ($B = -42.9$, 95% CI −77.5 to −8.3) spent less time in LPA than those with relapsing remitting MS. Walking capacity, assessed using the 12-item MS walking scale (MSWS-12), was associated with time in MVPA ($B = -0.36$, 95% CI −0.72 to −0.01).

Conclusion: This work identifies walking capacity and type of MS as correlates of PA, which may indicate development of interventions to promote PA.

Keywords: multiple sclerosis, light physical activity (LPA), moderate to vigorous physical activity (MVPA), physical activity, MS

INTRODUCTION

Increasing physical activity (PA) represents a safe (1) and cost-effective (2) approach for managing the sequelae of multiple sclerosis (MS) (3). PA is positively associated with walking mobility (4), quality of life (5), depression and fatigue (6), and cardiovascular health (7). Despite these benefits, physical inactivity is common. People with MS have lower step counts (8) and engage in significantly less moderate-to-vigorous physical activity (MVPA) than the general population (8, 9). Between 60% (10) and 80% of people with MS do not meet the minimum recommended PA volumes (9).

Identifying modifiable variables that are associated with PA may establish targets to support changes in PA and indicate the development of programmes to promote PA among people with MS (11). Further, establishing non-modifiable demographic and clinical correlates of PA may inform decisions on which subgroups of people with MS may particularly benefit from PA intervention. A systematic review by Streber et al. (12) identified that employment status, education level, disability level, walking limitations, and self-efficacy are consistently correlated with PA in people with MS. Age, BMI, falls history, type of MS, fatigue, and depression were inconsistently associated with PA (12). Inconsistent findings between studies may in part be attributable to limited consideration of possible confounding factors such as disability status. Further, the review identified that findings were limited by the use of self-report measures of PA (12). Studies published since the review continue to apply self-report PA measures (13–17). As self-report measures of PA demonstrate poor agreement with objective measures of PA such as accelerometers (18), employing objective measures is important to ensure accurate quantification of PA while examining its correlates.

This work, therefore aimed to address limitations of previous research by examining modifiable and non-modifiable correlates of objectively measured light PA (LPA) and MVPA in ambulatory adults with MS. A second aim was to examine if associations were modified by disability status, as measured by the expanded disability status scale (EDSS).

METHODS

This cross-sectional study presents an analysis of baseline data from the iStep-MS trial. The iStep-MS trial was a feasibility randomized controlled trial of a behavior change intervention, which aimed to increase PA and reduce sedentary behavior in people with MS (19).

Participants

Sixty people with MS were recruited from an MS Therapy Center in England and the MS Society UK website. Inclusion criteria were: a self-reported diagnosis of MS, self-reported relapse-free for the past 3 months, free of unstable medical conditions such as unstable angina that would make it unsafe to participate in PA, and ability to independently walk within the home with or without a walking aid. Exclusion criteria were pregnancy and ongoing participation in other trials. Written informed consent was obtained from all participants. The College of Health and Life Sciences Research Ethics Committee in Brunel University London (6181-NHS-Apr/2017-7016-2) approved this work.

Independent Variables

We included demographic and clinical characteristics as independent variables in this analysis. Participants completed a questionnaire at baseline that provided information on their age, sex, ethnicity, living arrangement (i.e., living alone or living with family/partner), employment status, marital status, type of MS, and duration of MS. Participants could request support from the researcher to complete the questionnaire if required. Participants were categorized as EDSS (20) levels 1.0–4.0 or 4.5–6.5. A researcher measured participants' weight using a Seca 875 Flat Scale, height using a Seca 213 portable stadiometer, and then calculated body mass index (BMI). Waist circumference was measured using a tape (Seca) on bare skin, to the nearest 0.1 cm midway between the lower rib margin and the iliac crest at the end of gentle expiration. The mean of two measurements was used for waist circumference.

Fatigue was assessed using the modified fatigue impact scale (MFIS). Self-efficacy was assessed using the multiple sclerosis self-efficacy scale (MSSE) function and control subscales. Walking capability was assessed using the 12-item MS walking scale (MSWS-12). The physical and psychological impact of MS was assessed using the multiple sclerosis impact scale (MSIS-29). Health-related quality of life (HRQOL) was assessed using EuroQol-5D-5L (21). The United Kingdom value set was used to calculate a utility score (22). Participation over four domains (autonomy indoors, family role, autonomy outdoors, and social life and relationships) was assessed using the impact on participation and autonomy questionnaire (IPA). The median score was obtained for each participant for each subscale. A full description of the measurement of these variables measured is provided elsewhere (23). Variable scoring is outlined in **Supplementary Table 1**.

Dependent Variables

Time in light PA and MVPA were included as dependent variables in this analysis. Participants were asked to wear an ActiGraph wGT3X-BT accelerometer (ActiGraph, Pensacola, Florida, USA) for 7 days. The ActiGraph was worn on an elastic belt at the mid-axillary line at the hip during waking hours only and removed for any water-based activities. Non-wear-time was defined as $\geq$90 consecutive minutes of 0 counts (24) and was validated against wear-time diaries kept by participants. Days with $\geq$10 h of wear data were considered valid and participants with at least 3 valid days were included in analysis (25). LPA was determined using a threshold of $\geq$100 counts per minute and $<$1,745 counts per minute. MVPA was classified as $\geq$1,745 counts per minute using established MS specific cut points (26).

Data Analysis

Statistical analysis was performed using Stata, version 16.0 (StataCorp LP, College Station, Texas). The distribution of data was examined using histograms, Q–Q plots, and cross-tabulations. Data are summarized as mean, standard deviation, median, minimum, maximum, frequencies, and proportions as appropriate. Separate linear regression models were used to examine unadjusted associations between demographic and clinical characteristics (included as independent variables), and LPA and MVPA (included as dependent variables), respectively. All demographic and clinical characteristics that were associated with LPA at the level of $p < 0.05$ were included in a multiple linear regression model. Similarly, characteristics associated with MVPA at the level of $p < 0.05$ were included in a multiple linear regression model. Where we observed an association between a characteristic and LPA or MVPA, we included an interaction term between the characteristic and EDSS category to examine if the association was modified by disability status. Assumptions of linear regression were assessed by visually inspecting Q–Q plots

TABLE 1 | Participant characteristics.

	n (%)	Mean (SD)	Median (range)
Age, years	58	56.8 (9.2)	57 (37-74)
Female	39 (67)		
Ethnicity			
White	51 (88)		
Black	4 (7)		
Asian	3 (5)		
Living arrangement			
Lives alone	8 (14)		
Lives with partner/spouse/family	50 (86)		
Employment status			
In paid employment	19 (33)		
Not in paid employment	39 (67)		
Marital status			
Married/partnered	45 (78)		
Not married/partnered	13 (22)		
BMI, kg.m^2	58	26.2 (5.5)	24.29 (16.77–47.99)
Waist circumference, cm	58	96.4 (15.2)	95.8 (70–154.2)
MS duration, years	57	15.4 (9.8)	13 (1–42)
Type of MS			
Relapsing-remitting	20 (35)		
Secondary progressive	21 (36)		
Primary progressive	13 (22)		
Unknown	4 (7)		
EDSS			
1.0–4.0	15 (26)		
4.5–6.5	43 (74)		
MFIS (0–84)	58	43.2 (18.4)	44 (1–81)
MMSE control (90–900)	58	573.3 (201.2)	570 (230–890)
MMSE function (90–900)	58	661.2 (197.6)	690 (180–900)
MSWS-12, %	58	74.6 (20.0)	79.2 (20.0–100.0)
MSIS-29 physical (0–100)	58	43.0 (21.3)	42.5 (3.8–86.3)
MSIS-29 psychological (0–100)	58	31.6 (20.1)	30.6 (0.0–86.1)
EQ-5D-5L utility	58	0.63 (0.19)	0.64 (-0.04 to 1.00)
IPA: autonomy indoors (0–4)	58	0.67 (0.87)	0 (0–3)
IPA: family role (0–4)	58	1.33 (0.94)	1 (0–3)
IPA: autonomy outdoors (0–4)	58	1.57 (1.11)	1 (0–4)
IPA: social life and relationships (0–4)	58	0.48 (0.60)	0 (0–2)

BMI, body mass index; EDSS, Expanded Disability Status Scale; IPA, Impact on Participation and Autonomy Questionnaire; MFIS, Modified Fatigue Impact Scale; MS, multiple sclerosis; MSSE, Multiple Sclerosis Self-Efficacy Scale; MSWS-12, Twelve Item MS Walking Scale; SD, standard deviation. MS duration n = 1 missing.

of residuals and scatter plots of residuals against fitted values. There was no evidence of heteroscedasticity or non-normally distributed residuals.

RESULTS

Two participants did not return the accelerometer, resulting in 58 participants included in the analysis. **Table 1** displays the demographic and clinical characteristics of included participants. Participants had a mean (standard deviation [SD]) age of 56.8 (9.2) years, and were predominantly women (67%) and white (88%). Most (86%) lived with a partner, spouse, or family member, and 14% lived alone. Seventy-eight per cent were married/partnered and 13% were not married/partnered. Sixty-seven per cent were not in paid employment, 33% were in paid employment. Most (74%) were in EDSS 4.5–6.5, and 26% were in EDSS 1.0–4.0. Approximately a third of participants had relapsing-remitting MS, 36% had secondary progressive MS, and 22% had primary progressive MS. Median duration since diagnosis of MS was 13 years (range 1–42 years).

Participants wore the accelerometer for a mean (SD) 6.77 (0.83) days and 851.20 (84.61) min/day (range 658–1092.16 min/day). Time spent in LPA and MVPA is described in **Table 2**.

Table 3 presents the unadjusted and adjusted associations between demographic and clinical characteristics, and time in LPA. In unadjusted analyses, women spent on average 34.6 min (95% CI 6.3–62.9 min) more than men in LPA per day. Asian participants spent on average 63.4 min (95% CI 2.4–124.3 min) less than white participants in LPA per day. People with secondary progressive and primary progressive MS spent less time in LPA than people with relapsing remitting MS (coeff. −56.6, 95% CI −85.9 to −27.4, and coeff. −53.5, 95% CI −86.8 to −20.1, respectively). The MMSE function subscale and EQ-5D-5L utility score were positively associated with time in LPA (coeff. 0.10, 95% CI 0.03–0.16, and coeff. 101.4, 95% CI 32.0–170.7, respectively). The MSIS-29 physical subscale was negatively associated with time in LPA (coeff. −0.68, 95% CI −1.32 to −0.05).

TABLE 2 | Time spent in light and moderate-to-vigorous activity ($n = 58$).

	Mean (SD)	Median (IQR)
Light PA (min/day)	157.74 (52.64)	159.12 (125.96–192.32)
Moderate-to-vigorous PA (min/day)	17.93 (16.21)	11.68 (5.07–29.14)

IQR, interquartile range; PA, physical activity; SD, standard deviation.

In the multiple linear regression model, only type of MS was associated with time in LPA. Specifically, people with secondary progressive MS spent on average 54 min (95% CI −84.7 to 23.3 min) less in LPA per day than those with relapsing remitting MS. People with primary progressive MS also spent on average 42.9 min (95% CI −77.5 to −8.3 min) less in LPA per day than people with relapsing remitting MS. There was no evidence that the association between type of MS and LPA differed in those with EDSS score 1.0–4.0 compared with those with EDSS score 4.5–6.5. When an MS type-by-EDSS interaction term was included in the multiple linear regression, there was no evidence that the association between type of MS and LPA was different between people with EDSS levels 1.0–4.0 and those in levels 4.5–6.5 ($p = 0.565$).

TABLE 3 | Associations between demographic, clinical characteristics, and light physical activity.

	LPA (min/day)		LPA (min/day)	
	Unadjusted Coeff. (95% CI)	*p*	Adjusted Coeff. (95% CI)	*p*
Age, years	−1.32 (−2.81 to 0.18)	0.084	-	
Female (ref: male)	34.6 (6.3 to 62.9)	0.017	20.27 (−6.84 to 47.38)	0.139
Ethnicity (ref: White)				
Black	−29.6 (−82.9 to 23.6)	0.270	−27.2 (−76.4 to 21.9)	0.271
Asian	−63.4 (−124.3 to −2.4)	0.042	−62.0 (−127.0 to 2.9)	0.061
Living alone (ref: living with partner/spouse/family)	−16.9 (−57.1 to 23.4)	0.405	-	
Not in paid employment (ref: in paid employment)	−20.0 (−49.3 to 9.3)	0.177	-	
Not married/partnered (ref: married/partnered)	−0.67 (−34.2 to 32.8)	0.968	-	
BMI, kg.m^2	−1.75 (−4.26 to 0.76)	0.168	-	
Waist circumference, cm	−0.62 (−1.53 to 0.29)	0.179	-	
MS duration, years	−1.22 (−2.64 to 0.20)	0.092	-	
Type of MS (ref: Relapsing-remitting)				
Secondary progressive	−56.6 (−85.9 to −27.4)	<0.000	−54.0 (−84.7 to −23.3)	0.001
Primary progressive	−53.5 (−86.8 to −20.1)	0.002	−42.9 (−77.5 to −8.3)	0.016
Unknown	−14.4 (−65.7 to 36.8)	0.575	−21.7 (−73.1 to 29.7)	0.400
EDSS 4.5-6.5 (ref: EDSS 1.0-4.0)	−25.1 (−56.3 to 6.1)	0.112	–	
MFIS (0–84)	−0.26 (−1.02 to 0.51)	0.506	-	
MMSE function (90–900)	0.10 (0.03 to 0.16)	0.005	−0.01 (−0.12 to 0.10)	0.849
MMSE control (90–900)	0.05 (−0.02 to 0.12)	0.125	-	
MSWS-12, %	−0.66 (−1.34 to 0.02)	0.057	-	
MSIS-29 psychological (0–100)	−0.32 (−1.02 to 0.37)	0.355	-	
MSIS-29 physical (0–100)	−0.68 (−1.32 to −0.05)	0.036	0.26 (−0.69 to 1.22)	0.583
EQ-5D-5L utility	101.4 (32.0 to 170.7)	0.005	76.8 (−17.2 to 170.7)	0.107
IPA: autonomy indoors (0–4)	−19.1 (−34.5 to −3.6)	0.017	6.88 (−14.2 to 27.9)	0.514
IPA: family role (0–4)	−12.4 (−26.9 to 2.2)	0.094	-	
IPA: autonomy outdoors (0–4)	−10.3 (−22.7 to 2.1)	0.103	-	
IPA: social life and relationships (0–4)	−29.9 (−52.0 to −7.8)	0.009	−16.6 (−42.5 to 9.3)	0.204

BMI, body mass index; EDSS, Expanded Disability Status Scale; IPA, Impact on Participation and Autonomy Questionnaire; MFIS, Modified Fatigue Impact Scale; MS, multiple sclerosis; MSSE, Multiple Sclerosis Self-Efficacy Scale; MSWS-12, Twelve Item MS Walking Scale; SD, standard deviation.

TABLE 4 | Associations between demographic, clinical characteristics, and moderate-to-vigorous physical activity.

	MVPA (min/day)		MVPA (min/day)	
	Unadjusted Coeff. (95% CI)	***p***	**Adjusted Coeff. (95% CI)**	***p***
Age, years	−0.59 (−1.04 to −0.14)	0.011	0.04 (−0.52 to 0.60)	0.887
Female (ref: male)	8.79 (−0.07 to 17.65)	0.052	-	
Ethnicity (ref: White)				
Black	−8.34 (−25.27 to 8.58)	0.327	-	
Asian	−8.25 (−27.63 to 11.11)	0.397	-	
Living alone (ref: living with partner/spouse/family)	−6.16 (−18.53 to 6.20)	0.322	-	
Not in paid employment (ref: in paid employment)	−16.9 (−24.8 to −8.9)	<0.000	−2.59 (−14.34 to 9.17)	0.659
Not married/partnered (ref: married/partnered)	−5.05 (−15.28 to 5.18)	0.327	-	
BMI, kg.m^2	0.09 (−0.70 to 0.87)	0.820	-	
Waist circumference, cm	−0.05 (−0.33 to 0.24)	0.731	-	
MS duration, years	−0.60 (−1.02 to −0.18)	0.006	−0.39 (−0.90 to 0.12)	0.126
Type of MS (ref: relapsing-remitting)				
Secondary progressive	−17.3 (−26.2 to −8.3)	<0.000	−6.63 (−16.95 to 3.69)	0.202
Primary progressive	−18.3 (−28.5 to −8.10)	0.001	−10.53 (−23.88 to 2.81)	0.118
Unknown	−8.0 (−23.6 to 7.7)	0.314	−6.56 (−24.94 to 11.83)	0.475
EDSS 4.5-6.5 (ref: EDSS 1.0-4.0)	−18.9 (−27.3 to −10.5)	<0.000	−5.23 (−16.82 to 6.36)	0.367
MFIS (0–84)	−0.27 (−0.50 to −0.05)	0.019	0.05 (−0.32 to 0.42)	0.778
MMSE function (90–900)	0.03 (0.01 to 0.05)	0.004	−0.01 (−0.05 to 0.02)	0.470
MMSE control (90–900)	0.02 (0.00 to 0.05)	0.020	0.00 (−0.04 to 0.04)	0.979
MSWS-12, %	−0.49 (−0.67 to −0.32)	<0.000	−0.36 (−0.72 to −0.01)	0.047
MSIS-29 psychological (0–100)	−0.18 (−0.39 to 0.03)	0.086	-	
MSIS-29 physical (0–100)	−0.33 (−0.51 to −0.15)	0.001	0.13 (−0.29 to 0.54)	0.549
EQ-5D-5L utility	28.9 (7.3 to 50.5)	0.010	20.1 (−11.9 to 52.0)	0.211
IPA: autonomy indoors (0–4)	−4.78 (−9.62 to 0.06)	0.053	-	
IPA: family role (0–4)	−5.07 (−9.47 to −0.68)	0.024	0.80 (−4.96 to 6.57)	0.780
IPA: autonomy outdoors (0–4)	−4.94 (−8.62 to −1.26)	0.009	−1.55 (−7.83 to 4.73)	0.621
IPA: social life and relationships (0–4)	−7.65 (−14.59 to −0.70)	0.031	−1.71 (−9.72 to 6.30)	0.668

BMI, body mass index; EDSS, Expanded Disability Status Scale; IPA, Impact on Participation and Autonomy Questionnaire; MFIS, Modified Fatigue Impact Scale; MS, multiple sclerosis; MSSE, Multiple Sclerosis Self-Efficacy Scale; MSWS-12, Twelve Item MS Walking Scale; SD, standard deviation.

Table 4 presents the associations between demographic and clinical characteristics and time in MVPA. In unadjusted analyses, women spent more time in MVPA than men (coeff. 8.79, 95% CI −0.07 to 17.65). People who were not in paid employment spent less time in MVPA than those in paid employment (coeff. −16.9, 95% CI −24.8 to −8.9). People with secondary progressive MS and primary progressive MS spent less time in MVPA than those with relapsing remitting MS (coeff. −17.3, 95% CI −26.2 to −8.3, and coeff. −18.3, 95% CI −28.5 to −8.10, respectively). People with EDSS 4.5–6.5 spent less time in MVPA than those with 1.0–4.0 (coeff. −18.9, 95% CI −27.3 to −10.5). Age (coeff. −0.59, 95% CI −1.04 to −0.14), MS duration (coeff. −0.60, 95% CI −1.02 to −0.18), MFIS (coeff. −0.49, 95% CI −0.67 to −0.32), MSWS-12 (coeff. −0.49, 95% CI −0.67 to −0.32), and MSIS-29 physical subscale (coeff. −0.33, 95% CI −0.51 to −0.15) were negatively associated with time in MVPA. MMSE function subscale (coeff. 0.03, 95% CI 0.01 to 0.05), MMSE control subscale (coeff. 0.02, 95% CI 0.00 to 0.05), and EQ-5D-5L utility score (coeff. 28.9, 95% CI 7.3 to 50.5) were positively associated with time in MVPA. The IPA family role subscale (coeff. −5.07, 95% CI −9.47 to −0.68), autonomy outdoors subscale (coeff. −4.94, 95% CI −8.62 to −1.26), and social life and relationship subscale (coeff. −7.65, 95% CI −14.59 to −0.70) were negatively associated with time in MVPA.

In the adjusted model, only MSWS-12 was associated with time in MVPA. A 1% increase in MSWS-12 was associated with, on average, a decrease of 0.36 min of MVPA per day (95% CI −0.72 to −0.01 min/day). There was evidence that the association between MSWS-12 and MVPA differed depending on EDSS score, as indicated by the *p*-value for the EDSS-by-MSWS interaction term ($p = 0.028$). For those with an EDSS score of between 1.0 and 4.0, a 1% increase in MSWS-12 was associated with a decrease of 0.57 min of MVPA per day (95% CI −0.95 to −0.18; $p = 0.005$). However, there was no association between MSWS-12 and MVPA for those with an EDSS score of between 4.5 and 6.5 (coeff. −0.03, 95% CI −0.48 to 0.42, $p = 0.878$).

DISCUSSION

This study examined modifiable and non-modifiable correlates of accelerometer-determined LPA and MVPA in a sample of adults with MS. In the adjusted regression model, type of MS was associated with LPA, and walking capability as measured by the MSWS-12 was associated with MVPA.

In line with previous research, inverse relationships between MS duration (9), disability status (27–29), age (30), and MVPA were demonstrated in the unadjusted analyses. Being a woman and White were associated with higher levels of LPA. Recent research demonstrated that men with MS exhibit higher levels of LPA than women (31, 32). However, a review concluded that sex is inconsistently associated with PA (12). Disagreement may be explained by differences between studies in terms of the PA construct examined, disability level, or personal characteristics (e.g., self-efficacy) of the sample (12).

Two studies have examined the association between PA and ethnicity among people with MS. One found no difference in objectively measured MVPA between White people and people from other ethnic backgrounds (9), and the second found a difference in self-reported PA between Black and White participants (33). To our knowledge, this is the first study to examine the association between LPA and ethnicity. The high proportion of White participants limits this finding and exploration of PA participation, and influences of PA among individuals with MS from Black and Asian ethnic backgrounds is warranted. Further, the relatively low proportion of Black and Asian participants does reflect existing exercise (34) and PA (9) literature in people with MS, which is predominately composed of White participants, and highlights a need to identify how to engage and promote inclusion of people with MS from other ethnic backgrounds in similar studies.

No non-modifiable factor remained associated with MVPA in adjusted analyses. Only type of MS remained associated with LPA in adjusted analyses. People with secondary progressive and primary progressive MS spent on average 54 and 43 min per day less in LPA, respectively, than those with relapsing–remitting MS, even after controlling for sex, ethnicity, self-efficacy for function, physical impact of MS, quality of life, and participation and autonomy. Type of MS has been shown to be associated with objectively measured step count when controlling for age, cane use, number of years since MS diagnosis, employment status, and type of MS (30). Interventions for changing PA behavior in people with MS have predominantly included ambulatory participants with relapsing–remitting MS (35). The present results emphasize the need to provide interventions that promote PA to people with progressive disease courses.

In terms of potentially modifiable factors, employment status and fatigue were negatively associated with MVPA in the unadjusted analyses. Our findings align with previous research that demonstrates that unemployment (9) and fatigue (17) are negatively associated with PA.

In this work, both the EQ-5D-5L utility score and the MSIS-29 physical subscale correlated significantly with time in LPA and MVPA. These findings support cross-sectional research that demonstrated a positive association between quality of life and objective PA (36) and longitudinal studies that demonstrate alterations in PA yield favorable changes in physical and psychological disease impact (36). The IPA subscales were negatively associated with MVPA and LPA. This aligns with previous research in people with MS, which demonstrated poorer autonomy and participation in those with lower aerobic capacity (37). Experiences of participation and autonomy appear to be closely associated with perceived quality of life and disease impact (38). Focusing on strategies to enhance quality of life like social support (39) and assessment, and modification of environmental barriers which have a large and negative effect on participation (38, 40) in people with MS may influence these factors, and, in turn, positively influence PA.

Self-efficacy for function (i.e., confidence in performing behaviors associated with engaging in daily living activities) was positively associated with LPA and MVPA, and self-efficacy for control (i.e., confidence to manage disease symptoms, reactions, and impact on daily activities) was associated with MVPA. Self-efficacy is a consistent positive correlate of PA (12). Comparison with existing research is difficult due to varied PA data collection methods, examination of associations using univariable analyses (27, 41), or analyses that control for a wide range of confounding variables from environmental factors (16) to social cognitive theory constructs (42). In this work the relationship between self-efficacy and PA may have been confounded by the inclusion of MSWS-12 in the model which is negatively associated with both self-efficacy (43) and PA (44).

Walking capacity was the only independent predictor of MVPA in the adjusted analyses. No potentially modifiable factors remained associated with LPA. The negative association between MSWS-12 and MVPA in the adjusted analysis reflects previous research which demonstrated that more severe walking impairment is associated with reduced step count after controlling for disease duration and severity (45). Walking capacity fluctuates regularly across the disease course, even in those with relatively stable disease (46). Targeting interventions to improve walking capacity through for example core stability and balance (47) may represent a mechanism to help improve PA.

In this work, EDSS score significantly moderated the relationship between walking ability and MVPA. Walking capacity was associated with PA for participants with EDSS score 1.0–4.0. In participants with EDSS score $>$ 4.5 no association between walking capacity and PA was demonstrated. It is possible that in participants with EDSS 1.0–4.0 there was sufficient variation in walking capacity and PA to show an association, whereas for people in EDSS 4.5–6.5 variation in walking capacity and PA was too limited to find an association. Strategies to improve walking capacity may be particularly beneficial for increasing PA in people with lower EDSS scores. However, interventions that focus on the types of activity other than walking, including resistance training and adapted exercise modalities such as electrical stimulation cycling (48), may promote more sustainable PA for individuals with higher levels of disability or mobility limitations.

Strengths and Limitations

This study addressed limitations of previous research by using an objective PA measure and including a more diverse representation of people with both relapsing–remitting and progressive MS, rather than relapsing–remitting only. However, most participants were women and White, which limits the generalisability of results. Furthermore, as participants were recruited from an MS Center and the MS Society website they may be more engaged with PA than the general MS population, and therefore more motivated to take part in PA.The cross-sectional nature of this research precludes any inferences of causality. Finally the small sample size is a limitation.

Implications

In summary, the findings of this study add to previous research that suggests age, sex, ethnicity, type of MS, duration of MS, and disability level are potentially non-modifiable predictors of PA (12). Similarly, in agreement with previous research, self-efficacy, fatigue, quality of life, employment status, participation, and autonomy may be important and potentially modifiable factors for modulating PA.

Although these findings suggest specific subgroups of people and potential modifiable factors to target to increase PA in this population, the majority of these factors were not associated with PA, when other non-modifiable and modifiable factors were controlled for. Therefore, although they are important to consider when developing and implementing PA interventions, they should not be considered in isolation. Walking capacity and the type of MS were the only independent correlates of PA. Exploring the barriers and facilitators to PA according to type of MS may inform development of PA interventions. Further, identifying strategies to improve walking capacity and supporting people with MS to engage in a variety of types of PA should be considered in future interventions to increase PA.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the College of Health and Life Sciences Research Ethics Committee in Brunel University London. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

JF and JR conceived the idea and analyzed the data. MN, AS, CK, GL, WH, CV, and LD contributed to the writing and assisted with the interpretation. JF completed this work while working at Brunel University London. All authors have read and approved the final manuscript.

FUNDING

This work was supported by the MS Society UK (grant number 53).

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fresc.2021.726436/full#supplementary-material

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Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers
in Rehabilitation Sciences

ORIGINAL RESEARCH
published: 11 January 2022
doi: 10.3389/fresc.2021.737384

Validity and Acceptability of Wearable Devices for Monitoring Step-Count and Activity Minutes Among People With Multiple Sclerosis

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Specialty section:
This article was submitted to Disability, Rehabilitation, and Inclusion, a section of the journal Frontiers in Rehabilitation Sciences

Received: 06 July 2021
Accepted: 19 November 2021
Published: 11 January 2022

Citation:
Lavelle G, Norris M, Flemming J, Harper J, Bradley J, Johnston H, Fortune J, Stennett A, Kilbride C and Ryan JM (2022) Validity and Acceptability of Wearable Devices for Monitoring Step-Count and Activity Minutes Among People With Multiple Sclerosis.
Front. Rehabilit. Sci. 2:737384.
doi: 10.3389/fresc.2021.737384

Multiple wearable devices that purport to measure physical activity are widely available to consumers. While they may support increases in physical activity among people with multiple sclerosis (MS) by providing feedback on their performance, there is little information about the validity and acceptability of these devices. Providing devices that are perceived as inaccurate and difficult to use may have negative consequences for people with MS, rather than supporting participation in physical activity. The aim of this study was, therefore, to assess the validity and acceptability of commercially available devices for monitoring step-count and activity time among people with MS. Nineteen ambulatory adults with MS [mean (SD) age 52.1 (11.9) years] participated in the study. Step-count was assessed using five commercially available devices (Fitbit Alta, Fitbit Zip, Garmin Vivofit 4, Yamax Digi Walker SW200, and Letscom monitor) and an activPAL3μ while completing nine everyday activities. Step-count was also manually counted. Time in light activity, moderate-to-vigorous activity, and total activity were measured during activities using an Actigraph GT3X accelerometer. Of the 19 participants who completed the validity study, fifteen of these people also wore the five commercially available devices for three consecutive days each, and participated in a semi-structured interview regarding their perception of the acceptability of the monitors. Mean percentage error for step-count ranged from 12.1% for the Yamax SW200 to −112.3% for the Letscom. Mean step-count as manually determined differed to mean step-count measured by the Fitbit Alta ($p = 0.002$), Garmin vivofit 4 ($p < 0.001$), Letscom ($p < 0.001$) and the research standard device, the activPAL3μ ($p < 0.001$). However, 95% limits of agreement were smallest for the activPAL3μ and largest for the Fitbit Alta. Median percentage error for activity minutes was 52.9% for the Letscom and 100% for the Garmin Vivofit 4 and Fitbit Alta compared to minutes in total activity. Three inductive themes were generated from participant accounts: *Interaction with device*; *The way the device looks and feels; Functionality.* In conclusion, commercially available devices demonstrated poor criterion validity when measuring step-count and activity time in people with MS. This negatively affected the acceptability of devices, with perceived inaccuracies causing distrust and frustration. Additional considerations when designing devices for people with MS include an appropriately sized and lit display and ease of attaching and charging devices.

Keywords: multiple sclerosis, wearable devices, physical activity, validity, acceptability, step-count

INTRODUCTION

Multiple sclerosis is a chronic, neurological condition affecting millions of people globally. It is estimated that a total of 2.8 million people live with a diagnosis of MS worldwide (35.9 per 100,000 population), with rising global prevalence rates observed since 2013 (1). Approximately 5,000 new cases of MS are diagnosed each year in the United Kingdom (UK), and it is more than two times as common in females than males (272 vs. 106 per 100,000 population) (2). A reduction in activity levels soon after diagnosis is common among people with MS, driving public health recommendations to help tackle these high levels of inactivity (3). Although physical activity may have several benefits for people with MS such as improved mental health, reduced fatigue, better walking performance, and lower mortality (4–7), many people with MS are inactive (8). Behavior change techniques, such as goal setting, providing feedback on performance, and self-monitoring of behavior, may support people with MS to increase physical activity (9). There is evidence from studies of the general population that monitoring physical activity alone results in an increase in physical activity (10). Multiple wearable devices that purport to measure physical activity are widely available to consumers. There is a large evidence base surrounding the validity of these devices to measure physical activity in the general population (11). Devices produced by Fitbit are, by far, the most frequently studied and appear to measure steps accurately, although validity may vary between devices (11). Gait deficits are a common feature of MS, and significant effects on gait even for those with relatively mild disability have been observed (12). This may affect the accuracy of these wearable devices when used by people with MS. However, few studies have specifically validated these devices in people with MS.

One study examined the criterion validity of five wearable devices and three smartphone applications for measuring steps among individuals with MS while walking on a treadmill (13). Devices included the Fitbit One and Fitbit Flex, Yamax SW200 Digi-walker, and Apple Health application. The Fitbit One demonstrated the best criterion validity for measuring steps, and measurement error was within 3% relative to manually counted steps (13), which is suggested as acceptable error (11). The Yamax SW200 and SW 401 were also compared against manually counted steps during treadmill walking in a group of adults with MS who could walk without an aid (14). Devices detected between 68.4 and 84.5% of observed steps during slow walking speeds but were more accurate during faster walking speeds, detecting between 95.6 and 100.5% of steps. Step-count from the Yamax SW200 over 7 days was also strongly correlated with step-count recorded by an Actigraph 7164 accelerometer (15, 16). Similarly, step-count recorded by the Fitbit Flex was strongly correlated with manually counted steps during overground walking and with steps recorded by an accelerometer over 7 days (17).

Although these studies provide some information on the validity of commercially available devices, most focus on one type of pedometer and not more recently developed wearables. Furthermore, criterion validity of these devices against manually counted steps was only assessed during walking. Despite being validated, adults with MS frequently perceived the Yamax SW200 to be inaccurate when monitoring their step-counts over 12 weeks (18). This suggests that, while these devices may be accurate for measuring step-count during walking in controlled environments, they are less accurate at measuring step-count during activities of daily life.

People with MS reported additional challenges with using the Yamax SW200 to monitor step-count, including difficulties attaching it, limited durability, and difficulties opening the device to view step-count on the digital display (18). These challenges resulted in frustration and had a negative impact on participants' motivation to increase step-count (18). These findings highlighted the importance of providing a device that is perceived as accurate and easy-to-use when asking individuals with MS to monitor physical activity. However, we were unable to identify any studies examining the acceptability of devices for monitoring physical activity among people with MS. Even if devices are valid, they will not be worn by people with MS and of little use for supporting physical activity behavior change, if they are not acceptable.

The aim of this study was, therefore, to assess the validity and acceptability of commercially available devices for monitoring step-count and activity time among people with MS.

MATERIALS AND METHODS

Design

A mixed methods trial design was carefully chosen to collect both qualitative and quantitative perspectives of each of the five commercially available devices. Combining data on both the validity of each device, in addition to capturing the views of people with MS on the perceived acceptability of each device, allows for a broader interpretation of results, with more clinically meaningful applicability of findings, given the inclusion of the participant voice. Mixed-methods design has been recommended as best practice in health outcomes research to enhance scientific rigor and ensure a focus on patient-led priorities (19). This study was conducted in two phases. In the first phase, we examined the criterion validity of five devices that we considered commercially available by comparing step-count against manually counted steps. We considered monitors to be commercially available if they can be purchased by anyone in order to monitor daily activity and do not require purchasing additional software to process activity data. We also compared step-count from a device commonly used in research, the activPAL3μ, against manually counted step-count, to allow us to comment on the criterion validity of commercially available devices relative to a research standard device. We further compared activity minutes obtained from the commercially available devices to time in light, moderate-to-vigorous and total activity from the Actigraph GT3x+. We used a cross-sectional design for Phase 1 of this study. That is, data were collected on one occasion during a 3-h session at Brunel University London. Following completion of Phase 1, the participants were asked to participate in the second phase of the study, to examine the acceptability of the commercially available devices. In the second phase, the participants wore each monitor for 3 days of everyday use and participated in a short face-to-face semi-structured interview at the end of each 3-day period. This design emulates "real-life" usage compared to the limitations imposed in controlled laboratory environments such as with the validity components of this work. The directed focus on various factors of acceptability and the aim to summarize commonalities between the participants positions this phase within a subtle realist approach.

Ethical approval was provided by Brunel University London's Research Ethics Committee (REC) and the Health Research Authority and Health and Care Research West Scotland (REC reference 18/WS/0161). The participants provided written informed consent to participate in each phase of the study.

Participants

The participants were recruited from an outpatient clinic at Hillingdon Hospital, MS support groups in the London Borough of Hillingdon, and a database of people with MS who previously consented to be contacted about research. Inclusion criteria were: a self-reported diagnosis of MS; over 18 years; relapse free for the past 3 months; able to independently walk with or without a walking aid within their home environment; free of unstable or acute medical conditions, e.g., unstable angina; and an ability to comprehend and follow all instructions relating to participation in the study. The participants were excluded if they were pregnant or participating in an alternative research study. The participants received a £20 voucher of their choice on completion of the study.

Procedures

The participant's self-reported age, height, weight, Expanded Disability Status Scale (EDSS) score, patient determined disease steps (PDDS), type of MS, duration of MS, and self-selected walking speed were recorded. Walking speed was measured as the average of three trials of walking 10 m overground in a straight corridor. The participants completed activities using a walking aid if required.

TABLE 1 | Description of activities.

Activity	Description
Deskwork	Participants sat at a desk, browsed the internet, made a phonecall, and typed for ten minutes.
Elevator	Participants travelled up and down three floors in an elevator.
Washing and drying dishes	Participants washed and dried dishes at a sink for ten minutes
Stairs	Participants ascended and descended one flight of stairs.
Indoor walking	Participants walked indoors along a straight 10 m walkway at a self-selected speed.
Walking with obstacles	Participants walked indoors at a self-selected speed navigating around objects on the ground.
Outdoor walking	Participants walked outdoors along a predefined route on a path that included straight paths, bends, and stepping on and off curbs, at a self-selected speed.
Stationary cycling	Participants cycled for 10 minutes at a self-selected speed on an upright cycle ergometer.
Driving	Participants who arrived to their appointment by car drove a predefined loop of the university campus at 20 mph

Phase 1: Validity

The participants performed eight activities in a controlled environment at Brunel University London while wearing the seven activity devices simultaneously. These were all worn according to recommended placement areas, including the wrist, hip/waist, and mid-thigh. Where more than one monitor was placed in the same area, e.g., wrist, steps were taken to ensure good contact and to limit inappropriate movement of the device. The participants could choose not to perform an activity if they believed they were unable to complete it or if there were any safety concerns. The participants rested in a seated position between each activity. The activities are described in **Table 1**.

Commercially Available Devices

We evaluated the following five commercially available devices: Fitbit Alta; Fitbit Zip; Garmin Vivofit 4; Yamax SW200 Digi-walker pedometer; and Letscom activity monitor. A description of each monitor is provided in **Supplemental Material**. We selected these devices as they vary in terms of (1) cost, (2) the type of data they collect, (3) how they are attached, (4) how data are displayed, and (5) the mode of charging. **Supplementary Table 1** also gives a full breakdown of device selection in terms of cost (ranging from £19.75 to £100), functionality, and feature of each device. We ensured to cover a range of devices in terms of these specific attributes (including cost, PA monitoring, display, clock function, attachment, mode of charging, and prompts to move). These were identified as important factors by people with MS when choosing a device to monitor physical activity (18). The Fitbit Alta, Garmin Vivofit 4, and Letscom activity monitor are wrist-worn devices that measure step-count and active minutes. The participants wore these devices on their right, or a least-affected arm. Wristbands were tightened to prevent movement of the monitor during activity. The Fitbit Zip and Yamax

SW200 Digi-walker pedometer are attached to the waistband of a person's clothes and monitor step-count. The participants wore these devices at their right, or a least-affected hip.

ActivPAL3μ

Steps were measured using the activPAL3μ activity monitor. The activPAL3μ is a small, lightweight device that is worn on the anterior aspect of the person's thigh. The participants wore the activPAL3μ on their right thigh. The activPAL3μ incorporates accelerometry and inclinometry data to provide information on step-count as well as the amount of time people spend on sedentary, upright, and ambulatory activities. Data were downloaded and processed using PAL software suite version 8.

Step Count

Steps were manually counted and measured during each activity using the five commercially available devices and the activPAL3μ. Steps were summed across each activity to provide total steps.

Step-count as displayed on the five commercially available devices at the start of the activity and at the end of the activity was recorded and subtracted to obtain steps measured during each activity. Additionally, the time that the activity started and stopped was recorded. Steps as measured by the activPAL3μ were calculated by extracting the corresponding time period from the activPAL3μ data and summing steps for this period. The participants were video-recorded, performing each activity, and steps for each activity were manually counted from video-recordings. Steps were counted by one individual. Accuracy of the step count by this rater was assessed by comparing it to steps counted by a second individual for 20% of the activities. Total steps across these activities were 1,802 for Rater 1 and 1,796 for Rater 2, an absolute difference of 6 steps or a difference of 0.3% relative to the first rater's step-count.

Activity Minutes

Activity minutes for the total period that activities were performed, from the start of the first activity to the end of the last activity, were identified from the display on the Fitbit Alta, Garmin Vivofit 4, and Letscom.

Minutes in light activity (LPA), minutes in moderate-to-vigorous activity (MVPA), and minutes in total activity were also obtained from the Actigraph GT3x+, which the participants wore on their right, or a least-affected side, at the hip during all activities. The Actigraph GT3x+ accelerometer is a small, lightweight triaxial accelerometer. Inbuilt sensors detect the magnitude of a person's acceleration in each plane, which is expressed as accelerometer counts per unit time. Data were collected in 1-s epochs.

Data were processed using the ActiLife 6 software. The time the first activity started and the time the last activity stopped was recorded. Accelerometer counts from the vertical axis were extracted for this time period. Accelerometer counts from the vertical axis only were used for data processing, because these were used to derive a cut point to classify MVPA in adults with MS (20). Minutes spent in MVPA were calculated by applying the cut point of 1,745 counts per minute, derived in a group of adults with MS, to the data (20). Light physical activity was identified as between 100 and 1,745 counts per minute. Minutes in total activity were calculated by summing time in LPA and time in MVPA.

Phase 2: Acceptability

The participants were asked to wear the five chosen commercially available devices over 15 days. They wore each monitor for three consecutive days. After the participants wore a monitor for 3 days, they participated in a brief semi-structured interview with the researcher regarding their perceptions of the acceptability of the monitor. A topic guide, developed from relevant literature and the aims of the study, was used to guide interviews. Questions included their experience of donning and doffing the monitor, process of using the device and seeing data, and perception of accuracy. The participants were then provided with the next monitor to wear for 3 days. The order in which the participants wore each monitor was randomized. After the participants wore the final monitor, they were asked an additional question about their preferred monitor and the comparable acceptability of the devices. Interviews were conducted in person at Brunel University London, in the participant's home, or in a location convenient for the participant. The interviews were audio-recorded and transcribed verbatim.

Data Analysis

The distribution of data was examined using appropriate graphs and tables. Mean, standard deviation, median, minimum, and maximum were used as appropriate to report participant characteristics, steps, and time in activity. We calculated the group percentage error for step-count as [(total steps from the device minus manually counted total steps)/manually counted total steps]*100 to allow comparison with other studies (11). We also report the number of the participants with a percentage error ≥ 5, ≥ 10, and $\geq 25\%$. Total steps from each device and manually counted steps were compared using paired *t*-tests. Bland-Altman plots were produced with 95% limits of agreement to compare agreement between each device and manually counted steps. We additionally calculated group percentage error for activity minutes for each device and compared activity minutes using Wilcoxon-signed rank tests. As it was unclear from the device manuals if activity minutes related to minutes in LPA, MVPA or LPA, and MVPA combined, we compared activity minutes from each device to minutes in LPA, MVPA, and total activity from the Actigraph GT3X. We did not calculate Bland-Altman plots for activity minutes as difference in activity minutes was not normally distributed. All analyses will be conducted using Stata version 14.0 (Statcorp, USA).

All interview recordings were transcribed and underwent framework analysis (21) by the same researcher. This method of analysis provides a clear audit trail of the analytical process, which enhances transparency. The technique involves five iterative stages of analysis; familiarization; identifying initial thematic framework through detailed line-by-line descriptive coding of the first five transcripts; labeling through which further minor adjustments were made to the framework; charting; mapping; and interpretation, following which significant themes can be presented. Labeling and charting included both deductive

TABLE 2 | Participant characteristics.

	n (%)	Mean (SD)	Median (range)
Age, yr	19	52.1 (11.9)	55 (27, 72)
Women	13 (68.4)		
BMI, kg.m^{-2}	19	29.4 (9.3)	26.3 (16.9, 52.2)
Type of MS			
Relapsing-remitting	16 (84.2)		
Secondary progressive	2 (10.5)		
Benign	1 (5.3)		
Relapse in past 3–12 months	5 (26.3)		
MS duration, yr	19	14.4 (11.8)	13 (0, 48)
EDSS			
1.0–4.0	10 (52.6)		
4.5–5.0	9 (47.4)		
Mobility aid use over 5m			
No aid	12 (63.2)		
Sticks or crutches	2 (10.5)		
Combination of sticks/crutches and wheelchair	5 (26.3)		1 (0, 5)
PDDS	19		1 (0, 5)
Walking speed, m/s	19	0.98 (0.36)	0.85 (0.64, 2.18)

codes such as comfort of a device as well as inductive codes that arose from the participant narratives. Initial coding, labeling, and thematic development were discussed in detail with another researcher who was familiar with the data and who had independently coded three transcripts.

RESULTS

Phase 1: Validity

Nineteen adults with MS were recruited to the study. Participant characteristics are presented in **Table 2**. The participants had a mean age of 52 years, ranging from 27 to 72 years. The majority were female with relapsing-remitting MS.

One participant did not complete the walking with obstacles activity, two people did not complete the cycling activity, three people did not complete the outdoor walking activity, three people did not complete the stairs activity, and five people did not complete the driving activity. Additionally, outdoor walking was not video-recorded for one person. Steps for each activity as measured by each device and as manually counted are presented in **Table 3**.

Percentage errors for each device are reported in **Table 4**. Percentage error was smallest for the Yamax SW200, although the Fitbit Zip had a similar percentage error. The Yamax SW200 and Fitbit Zip also had the fewest number of people with an error of >25%. Although the error for the research standard device, the activPAL3μ, was >25% for all the participants, the range was narrowest for the activPAL3μ. All devices except for the research standard device, the activPAL3μ, both overestimated and underestimated steps. The research standard device, the activPAL3μ, consistently underestimated steps by between 54 and 70%.

According to manually counted steps, all the participants had zero steps during deskwork, cycling, and driving. The research standard device, the activPAL3μ, also recorded zero steps for all the participants during these activities. The Fitbit Alta recorded >0 steps for four participants (21.1%) during deskwork, 13 participants (76.5%) during cycling, and 13 participants (92.9%) during driving. The Fitbit Zip recorded zero steps during deskwork. However, it recorded >0 counts for five participants (29.4%) during cycling and seven participants (50%) during driving. The Garmin Vivofit 4 recorded >0 steps for six participants (31.6%) during deskwork, eight participants (47.1%) during cycling, and all participants ($n = 14$) during driving. The Yamax SW200 Digi-walker pedometer recorded >0 steps for seven participants (36.8%) during deskwork, nine participants (52.9%) during cycling, and all the participants ($n = 14$) during driving. The Letscom monitor recorded >0 steps for one participant (5.3%) during deskwork, five participants (29.4%) during cycling, and five participants (35.7%) during driving.

The mean difference in total steps between manually counted steps and each monitor is described in **Table 5**. There was evidence that mean total steps differed between manually counted steps and the Fitbit Alta ($p = 0.002$), Garmin Vivofit 4 ($p < 0.001$), Letscom ($p < 0.001$), and research standard device, the activPAL3μ ($p < 0.001$). However, 95% limits of agreement were narrowest for the research standard device, the activPAL3μ, followed by the Garmin Vivofit 4 (**Table 5**). Limits of agreement were largest for the Fitbit Alta. However, they were similar for the Fitbit Zip.

Activity minutes reported by the Letscom, Fitbit Alta, and Garmin Vivofit 4, and minutes in LPA, MVPA, and total activity measured by the Actigraph GT3x+ are provided in **Table 6**. The Fitbit Alta recorded 0 activity minutes for 17 participants (89.5%), and the Garmin Vivofit 4 recorded 0 activity minutes for all 19 participants (100%). The Letscom did not record 0 activity minutes for any participant. No participant had 0 min in LPA or total activity as measured by the Actigraph GT3X. Six participants (33.3%) had 0 min in MVPA. Of these participants, all six had 0 activity minutes recorded by the Garmin Vivofit 4, five had 0 min recorded by the Fitbit Alta, and none had 0 min recorded by the Letscom. In comparison to min in total activity, median error was 52.9% for the Letscom and 100% for the Garmin Vivofit 4 and Fitbit Alta (**Table 7**). In comparison to minutes in LPA, median error was 48.6% for the Letscom and 100 for the Garmin Vivofit 4 and Fitbit Alta. There was a difference between activity minutes from each device and minutes in LPA and total activity (**Table 7**). There was also a difference between activity minutes and minutes in MVPA for the Letscom and Garmin Vivofit 4, but not for the Fitbit Alta ($p = 0.052$).

Phase 2: Acceptability

All 19 participants were invited to participate in Phase 2 of the study. Fifteen agreed and provided written informed consent to participate. Three inductive themes were generated from participant accounts: *Interaction with device*; *The way the*

TABLE 3 | Description of steps measured by each device and manually counted for each activity.

	Fitbit Alta	Fitbit Zip	Garmin Vivofit 4	Yamax SW200 Digi-Walker	Letscom	activPAL3μ	Manual count
Deskwork							
Median (min, max)	0 (0, 19)	0 (0, 0)	0 (0, 15)	0 (0, 6)	0 (0, 11)	0 (0, 0)	0 (0, 0)
Elevator							
Mean (SD)	10.2 (6.7)	12.1 (6.5)	11.6 (10.8)	12.7 (8.4)	–1.3 (5.7)	6.1 (3.0)	22.9 (7.6)
Median (min, max)	11 (0, 25)	14 (0, 21)	11 (0, 30)	14 (0, 21)	0 (-25, 0)	5.5 (3, 14)	21 (14, 48)
Washing dishes							
Mean (SD)	169.8 (125.6)	15.4 (13.4)	143.4 (71.6)	19.0 (13.4)	406.6 (255.2)	7.4 (4.5)	54.1 (29.0)
Median (min, max)	130 (24, 504)	13 (6, 67)	130 (59, 315)	15 (4, 57)	307 (0, 840)	5.5 (2, 19)	40 (20, 131)
Stairs[a]							
Mean (SD)	26.2 (11.3)	28.4 (8.2)	28.3 (15.3)	26.1 (8.2)	18.8 (19.6)	13.9 (2.8)	32.4 (4.6)
Median (min, max)	30 (0, 41)	31 (0, 36)	34 (0, 45)	31 (0, 36)	16 (0, 43)	14 (10, 20)	31.5 (27, 45)
Indoor walking							
Mean (SD)	32.6 (26.2)	37.0 (7.2)	37.5 (18.1)	36.6 (8.8)	32.3 (23.3)	18.4 (6.5)	40.3 (12.7)
Median (min, max)	33 (-62, 76)	36 (18, 56)	39 (0, 78)	36 (25, 67)	37 (0, 86)	16 (13, 35)	36 (28, 77)
Outdoor walking[b]							
Mean (SD)	274.4 (72.9)	254.4 (50.2)	280.8 (109.4)	182.6 (99.4)	271.4 (81.7)	137.5 (45.7)	304.1 (187.0)
Median (min, max)	258.5 (219, 530)	256 (119, 374)	263 (144, 667)	236 (0, 286)	265.5 (152, 552)	128 (107, 287)	262 (218, 976)
Cycling[c]							
Median (min, max)	81 (−9, 642)	0 (0, 544)	0 (0, 777)	1 (0, 154)	0 (0, 480)	0 (0, 0)	0 (0, 0)
Driving[d]							
Median (min, max)	29.5 (0, 144)	2 (0, 14)	112 (19, 236)	40 (1, 66)	0 (-29, 99)	0 (0, 0)	0 (0, 0)
Obstacles[e]							
Mean (SD)	47.9 (25.1)	47.3 (9.3)	61.3 (28.6)	42.1 (14.7)	57.0 (26.7)	26.2 (10.3)	61.1 (44.8)
Median (min, max)	43 (11, 123)	49.5 (19, 59)	55 (0, 130)	42.5 (16, 80)	48.5 (0, 120)	23, (19, 56)	48 (8, 222)
Total							
Mean (SD)	745.3 (357.8)	448.6 (249.7)	693.5 (289.6)	323.1 (118.7)	832.5 (314.7)	179.6 (83.7)	442.5 (269.6)
Median (min, max)	745 (147, 1425)	409 (83, 956)	730 (190, 1259)	365 (91,491)	965 (316, 1378)	190.5 (45, 428)	450 (101, 1442)

n = 19 for all devices except for activPAL3μ (n = 16) unless stated otherwise; [a]n = 16 for all devices except for activPAL3μ (n = 13); [b]n = 16 for all devices except for activPAL3μ (n = 13) and manual count (n = 15); [c]n = 17 for all devices except for activPAL3μ (n = 15); [d]n = 14 for all devices except for activPAL3μ (n = 13); [e]n = 18 for all devices except for activPAL3μ (n = 15).

TABLE 4 | Percentage error between visually counted steps and steps from each monitor.

Device	% Error, mean (95% CI)[a]	% Error, minimum, maximum (range)	≥ 5% Error, *n* (%)	≥10% Error, *n* (%)	≥25% Error, *n* (%)
Fitbit Alta	−80.6 (−113.0, −48.3)	−216.7, 48.3 (265.0)	19 (100)	18 (95)	18 (95)
Fitbit Zip	−14.9 (−47.0, 17.2)	−151.6, 82.8 (234.4)	17 (89)	14 (74)	8 (42)
Garmin Vivofit 4	−67.6 (−90.9, −44.2)	−168.4, 28.8 (197.2)	18 (95)	18 (95)	16 (84)
Yamax SW200	12.1 (−5.2, 29.4)	−79.5, 93.7 (173.2)	16 (84)	13 (68)	7 (37)
Letscom	−112.3 (−148,8, −75.9)	−318.8, 32.7 (351.5)	19 (100)	19 (100)	18 (95)
activPAL3μ[b]	59.0 (56.6, 61.3)	54.2, 70.3 (16.1)	19 (100)	19 (100)	19 (100)

[a]Positive value indicates the device underestimates steps in comparison to manual count.

[b]n = 16.

device looks and feels; Functionality. These are described below, illustrated through anonymised quotations.

Interactions With Device

This theme focuses on the accessibility of the device for individual use in which the qualities of the display and ease of data retrieval and charging were the key. A frequently noted feature was the importance of visibility with the participants preferring screens that were well-sized (noted 10 times), well-lit (five references), and easy to read (14 references). The Letscom received the highest number of positive comments in this regard:

> "*Display Is Clear, White on Black. Even in low Light, Is Easy to see." (Letscom-Participant 14).*
> *"Nice big time and date, heart rate easy to read." (Letscom-Participant 13).*

The importance of these features was noted by their absence or inadequacy (13 references to inadequate size, 13 references to inadequate or absent lighting, and 19 references to excessive complexity of display). The GarminVivofit 4 received 32 concerns, nearly double the number of any other device.

> *"Display is too small and not clear in different things to monitor. I don't understand the icons. It's not bright enough." (Garmin-Participant 2).*
> *"Don't like it. Small and compact… Screen too dark to read and not very clear with icons - difficult with vision problems." (Garmin-Participant 3).*

Of note here is the specific reference to potential visual problems, which are common in people with MS.

All the participants tried to retrieve data from the devices; nine people tried to retrieve data from the associated applications, and 13 people from the device itself. While 12 people noted that they had no difficulty with retrieving data, with the Letscom most commonly being noted as the easiest with eight references, challenges were raised with synching data between devices. The GarminVivofit 4 received the majority of negative comments (six references). This resulted in frustration and an inability to monitor their activity appropriately.

> *"syncing with phone is a nightmare. Does not self-sync and then does not record. Really aggravating to use. Needs auto-synch." (Garmin-Participant 12).*
> *"seems to underestimate steps in the app - what was on the watch and what was on the app was different (2,000 steps). So syncing issue." (Garmin-Participant 2).*

Further frustration came when charging the device was problematic. Only two of the devices used in this study required to be charged, the Letscom and Fitbit Alta. The majority of the participants (10 out of 11) that charged the Alta described the process as simple. In comparison, a large proportion of the participants who attempted to charge the Letsom (9 out of 10 participants) commented that this was a difficult process. The participant accounts detailed that this was due to the stiffness of the device and concern over causing damage. In response to the question "did you charge the Letscom?," one participant stated:

> *"yes, with major problems - nightmare. Need to wiggle and concerned with breaking charging port. Not easy at all. Husband also struggled. Will be a problem for anyone with hand problems." (Letscom-Participant 3).*

The reference to the challenge for people with dexterity issues is particularly relevant, given the prevalence of alterations in sensation and fine motor control in people with MS.

The Way the Device Looks and Feels

This theme draws together participant responses to both the aesthetic and comfort of the device. The participants were asked to score the comfort of devices out of 10, alongside feedback. The aesthetic of the device was not a component included in the topic guide but was noted as an important factor by many participants.

The Fitbit Zip, Fitbit Alta, and Yamax SW200 Digi-walker pedometer were generally seen as the most comfortable devices (mean scores of 9.3, 8.7, and 8.1, respectively). In the case of the Zip and Yamax SW200 Digi-walker pedometer, this appeared to relate to their positioning on the hip as the participants reported being fundamentally unaware of its presence once donned:

> *"Very comfortable 8/10. Didn't know I had it on." (Fitbit Zip-Participant 1).*
> *"10/10 - comfortable, tight fit, and stayed put, forget you're wearing it" (Fitbit Zip-Participant 10).*

All devices had some less positive comments on comfort. These related to the device catching on clothes or objects (Fitbit Zip, Letscom, and Yamax SW200 Digi-walker pedometer), the material of the product (all monitors), and irritation of the skin (Fitbit Alta, GarminVivofit, 4 and Letscom). However, these comments were limited, and, overall, all devices were considered acceptably comfortable (mean for each device range 7.1–9.3/10).

Many participants emphasized the importance of the aesthetic of the device (22 comments), requiring it not just to be fashionable, but to look sleek, modern, and up-to-date**:**

> *"It is comfortable and looks fashionable" (Letscom-Participant 6).*
> *"Also liked having a watch on the screen. Neat and tidy little thing" (Fitbit Alta-Participant 7).*

In contrast, devices were rejected if they disturbed the look of an outfit, usually through unwanted hip bulges. In general, female participants appeared to hold more importance than male in the aesthetic and design of the device (20/22 comments were from female participants). Additionally, several female participants commented on external judgement of the aesthetic, with colleagues or partners contributing to their positive or negative opinions of the device.

Functionality

When considering the functionality of the devices, the participants highlighted several relevant features, including perceived accuracy, the capacity of the device to encourage more

TABLE 5 | Difference and agreement between visually counted steps and steps from each monitor.

Monitor	Mean difference (95% CI), steps	*p*-value	95% Limits of agreement, steps
Fitbit Alta	−302.84 (-483.32, −122.36)	0.002	−1036.8, 431.1
Fitbit Zip	−6.16 (−181.05, 168.73)	0.942	−717.4, 705.0
Garmin Vivofit 4	−251.05 (-375.15, −126.95)	<0.001	−755.7, 253.6
Yamax SW200 Digi-walker pedometer	119.37 (−32.46, 271.19)	0.116	−498.0, 736.8
Letscom	−390.00 (−541.65, −238.35)	<0.001	−1006.7, 226.7
activPAL3μ[a]	277.31 (165.72, 388.90)	<0.001	−133.1, 687.8

Mean difference calculated as manually counted total steps minus monitor total steps.

[a] $n = 16$.

TABLE 6 | Description of activity time measured by Fitbit Alta, Garmin Vivofit 4, Letscom, and Actigraph GT3x.

Monitor		Activity time		Light activity		Moderate-to-vigorous activity		Total activity	
		Mean (SD)	Median (min, max)	Mean (SD)	Median (min, max)	Mean (SD)	Median (min, max)	Mean (SD)	Median (min, max)
Fitbit Alta	19	3.0 (10.2)	0 (0, 43)	n/a	n/a	n/a	n/a	n/a	n/a
Garmin Vivofit 4	19	0 (0)	0 (0, 0)	n/a	n/a	n/a	n/a	n/a	n/a
Letscom	18	18.7 (5.3)	18.5 (8, 31)	n/a	n/a	n/a	n/a	n/a	n/a
Actigraph GT3x+	18	n/a	n/a	34.6 (11.7)	31.5 (17, 56)	3.2 (3.2)	3 (0, 11)	37.7 (12.1)	35 (17, 57)

TABLE 7 | Percentage error, mean difference, and 95% limits of agreement between total activity measured by Actigraph GT3x and activity minutes from each monitor.

Device	*n*	% error, Median[a]	% error, Minimum, maximum (range)	≥ 5% error, *n* (%)	≥ 10% error, *n* (%)	≥ 25% error, *n* (%)	*p*-value[b]
Total activity							
Letscom	17	52.9	5.6, 65.1 (59.6)	17 (100)	16 (94.1)	15 (88.2)	<0.001
Fitbit Alta	18	100.0	−38.7, 100.0 (138.7)	18 (100)	18 (100)	18 (100)	<0.001
Garmin Vivofit 4	18	100.0	100.0, 100.0 (0.0)	18 (100)	18 (100)	18 (100)	<0.001
Light physical activity							
Letscom	17	48.6	5.3, 63.3 (58.1)	17 (100)	15 (88.2)	14 (82.3)	<0.001
Fitbit Alta	18	100.0	−38.7, 100.0 (138.7)	18 (100)	18 (100)	18 (100)	<0.001
Garmin Vivofit 4	18	100.0	100.0, 100.0 (0.0)	18 (100)	18 (100)	18 (100)	<0.001
Moderate to vigorous physical activity[c]							
Letscom		n/a	n/a	n/a	n/a	n/a	<0.001
Fitbit Alta		n/a	n/a	n/a	n/a	n/a	0.052
Garmin Vivofit 4		n/a	n/a	n/a	n/a	n/a	<0.001

[a] *Positive value indicates the device underestimates steps in comparison to Actigraph GT3x*

[b] *p value obtained from Wilcoxon signed rank test comparing activity time from each device to Actigraph GT3X.*

[c] *Unable to calculate percentage error because some people had 0 minutes of moderate-to-vigorous physical activity as measured by the Actigraph GT3X.*

physical activity, and the ease of donning and doffing the device and its stability once on.

The perceived accuracy of the data collected by the device was pivotal to its acceptability. These perceptions were based on the participants' self-monitored activity levels, and, in some cases, compared directly to devices already owned. While a range of positive comments were noted (32 comments across different devices), concerns predominated (45 comments). These were particularly noted with the YamaxSW200 Digi-walker pedometer (15 references), but also the Garmin Vivofit 4 (9 references) and Letscom (nine references). Perceived inaccuracy was an unacceptable quality in the device and led them to have strong negative views:

> *"If someone bought this thinking it was gonna help, especially someone with a medical condition, and it says, oh you've done these many steps, that's probably not great because if it's not consistent, then you can't see a consistent change……..It could be misleading if someone was really trying to look after their health and fitness or improve their step count, and that makes me kind of cross!" (Letscom-Participant 10).*
> *"Make a note, it's lying. I discovered, ha haha. I'm still sitting on the sofa here, and by moving back is one step. Liar, you're a liar. You can't even trust a funny ticky walk I had anymore. You are out of my life; I don't want you in my life. I thought I could love you, but I don't." (Yamax-Participant 6).*

As noted in the first quote, monitoring activity was expected to help the individual and over 50% of the participants discussed motivational elements of the device as unprompted comments. Positive motivational elements in the devices such as the ability to set goals, and prompts to move were valued.

> *"Yeah. Handy to see how active you are. Like to see steps as know goal is 10,000." (Fitbit Alta-Participant 11).*
> *"Gets a sense of achievement by doing more." (Fitbit Alta-Participant 6).*

The different devices had varying levels of monitoring and output. Accounts from the participants highlighted that levels of functionality of the device and accompanying app were a factor of acceptability. Where some were happy with basic functionality provided; others described wanting and liking a more holistic approach:

> *"Good info on the App-nutrition, trends, drinking water. After looking at how much water I should be drinking, I bought myself a water cup with measurements on it." (Fitbit Zip-Participant 13).*
> *"Would prefer something that measures heart rate, as can see patterns in my stress levels. From experience, I use heart rate levels on my own device to monitor when I need to rest." (Garmin-Participant 10).*

In contrast, several participants also described too much functionality as a negative component of a device, which, at times, could become off-putting or even de-motivating.

The ability to don and doff the device and ease of doing so were direct questions to all the participants for all devices. As with the ability to charge the device, the responses illuminate challenges in the required levels of dexterity, sensory integration, and strength:

> *"Trying to get it onto the trousers was fiddly. Pins and needles make it difficult. I put it on before the trousers went on." (Fitbit Zip-Participant 7).*
> *"It was stiff which was quite tricky - rubberised case, the spring is too strong. I needed both hands." (Fitbit Zip-Participant 3).*
> *"Hated it. Very difficult. Can't do the catch, its fiddly." (Yamax-Participant 13).*

Problems in this regard were almost exclusively noted with the devices that attached to clothes at a hip level.

DISCUSSION

This study aimed to assess the validity and acceptability of commercially available devices for monitoring step-count and activity time among people with MS. Of the commercially available monitors, the Garmin Vivofit 4 demonstrated the best agreement with manually counted steps. However, the mean step-count from the Garmin Vivofit differed to the mean manually counted steps, and the limits of agreement were wide, suggesting poor agreement and likely inaccurate measurement of step-count for individuals with MS. The research standard device, the activPAL3μ, performed best at measuring step-count in individuals with MS. All monitors, including the activPAL3μ, provided poor estimates of time in activity.

Percentage error was smallest for the Yamax SW200 and largest for the Letscom when compared to manually counted steps. However, percentage error for all monitors was high in comparison to that reported in a review of the validity of commercially available wearable devices among adults without mobility limitations and/or chronic diseases (133 studies) and without these conditions (36 studies) (11). Of 805 comparisons between devices and a criterion measure, 45.2% were within ± 3% measurement error, 42.7% were below −3% measurement error, and 12.1% were above 3% measurement error (11). This suggests that these devices perform particularly poorly in adults with MS. However, differences could also be due to the procedure used to validate the device and the type of the device.

Of the devices examined, only the Yamax SW200 had been validated against manually counted steps in people with MS. The Yamax SW200 detected between 68.4 and 100.5% of steps during treadmill walking at various speeds (14). Another study found a similar percentage error for the Yamax SW200 (8.5% compared to 12.1% in this study) (13). However, the proportion of the participants with ≥5, ≥10, and ≥25% error was much higher in our study than in the previous study (84 vs. 24%; 68 vs. 20%; and 37 vs. 11%) (13). This may be because we assessed criterion validity during a range of activities of daily living, while the previous study assessed criterion validity during two 500-m walking trials at a comfortable speed on a treadmill. Although these specific devices have not been evaluated in people with MS, four wearable motion sensors and three smartphone applications were evaluated in addition to the Yamax SW200 during treadmill walking (13). Percentage error for other devices ranged from 1.9

to 14.2% (13), with the Fitbit One demonstrating best absolute and relative precision, and fewest participants with ≥5% error (13). The higher percentage error we observed in this study for all monitors may be because they were evaluated during a range of activities and not just walking. Performance of the device likely differs, depending on the activity performed, which highlights the importance of assessing validity during everyday activities in order for findings to be more applicable to free-living conditions. One study compared daily step-count measured from the Fitbit Flex against daily step-count from an Actigraph accelerometer and observed a strong correlation but a difference in mean daily step-count between devices (17). Similarly, strong correlations were observed between the Yamax SW200 and Actigraph 7164 in free-living settings (15, 16). Correlation, however, does not equate to agreement (22).

All devices in this study had at least a 5% error for 84% or more of the participants compared to manually counted steps, with the research standard device, the activPAL3μ, having at least 25% error for all the participants. However, examining percentage error alone disguises the variability in individual error. The range of error for the research standard device, the activPAL3μ, was much smaller than for any other monitor at 16.1%. The range of error for other monitors was between 173.2% for the Yamax SW200 and 351.5% for the Letscom. This is reflected in the 95% limits of agreement, with the research standard device, the activPAL3μ, having the narrowest limits of agreement despite the mean step-count recorded by the activPAL3μ, being different to mean manually counted steps. The research standard device, the activPAL3μ, also consistently underestimated step-count, while other monitors under- and overestimated step-count. The lack of consistent direction in disagreement with manually counted steps for individuals makes it difficult to predict or account for error in these devices. Of the commercially available devices, the Garmin Vivofit 4 and Yamax SW200 demonstrated best agreement with manually counted steps. They also had the smallest range of error, albeit 197.2 and 173.2%. However, the limits of agreement were wide, particularly when compared to the mean total step-count. During a period of activity that results in a mean of 443 steps, the Garmin Vivofit 4 may overestimate steps by up to 756 steps and underestimate steps by up to 254 steps.

Of the three devices that provided activity minutes, it was not clear from the device manuals if activity minutes related to minutes in LPA, MVPA, LPA, and MVPA combined, or some other quantity. We, therefore, compared activity minutes from each device to LPA, MVPA, and total activity. However, activity minutes from all devices were not a good estimate of any measure of PA. Although the Letscom demonstrated the smallest median error compared to both total activity and LPA, the percentage error ranged from 5.6 to 65.1% for total activity and 5.3 to 63.3% for LPA. There was no evidence that activity minutes from the Fitbit Alta differed to minutes in MVPA. However, the Fitbit Alta recorded 0 min for 17 of the 19 participants, when only six participants had 0 min in MVPA, and none had 0 min in LPA or total activity. In addition, one of the two participants that the Fitbit Alta recorded > 0 activity minutes for did, in fact, have 0 min in MVPA. Overall, these data suggest none of these monitors should be used to estimate time in activity for people with MS.

No device was clearly preferred by the participants. However, four basic requirements were identified as being important for acceptability. These were being wrist worn (based on easier attachment and visibility), clear display, perceived accuracy, and offering something more than just step count. The latter is rooted in personal preference, with some participants preferring basic functionality, while others wanting more advanced monitoring features. Therefore, optional add-ons should be available to enhance acceptability for all users. Similarly, although aesthetics was important to many participants, requirements in terms of aesthetics differed between the participants.

Letscom was a clear favorite in terms of display characteristics and ease of data retrieval. The Garmin Vivofit 4 elicited the most negative reactions when the participants shared their thoughts on the device interface, as well as identifying difficulties synching the device. Similar issues were previously identified when exploring adults with MS experiences of monitoring step-count using the Yamax SW200 (18). However, this study highlights that these issues are not unique to the Yamax SW200. The strong emphasis on ease of attachment and charging and the visual display directly relate to additional physical challenges that people with MS may experience (23), and strongly indicate the need for manufacturers to consider the accessibility of their devices for people with impairments. Impaired vision and manual dexterity are not unique to MS and, in fact, may be experienced by many people as they age. Manufacturers need to consider designing products that are accessible to all. This has potential benefits to all users and not just those with specific conditions.

Perceived accuracy of the device was the overriding integral element to acceptability among people with MS. The participants particularly perceived the Yamax SW200, Letscom, and Garmin Vivofit 4 as inaccurate, despite the Garmin Vivofit 4 and the Yamax SW200 demonstrating best agreement with manually counted steps. We similarly identified that people with MS perceived the Yamax SW200 to be inaccurate when using it to monitor step-count over 12 weeks (18). This perception may be partly caused by these monitors recording steps during inactivity, which would have been particularly noticeable to participants when monitoring their activity at home. We found that the Yamax SW200 and Garmin Vivofit 4 incorrectly recorded steps during deskwork for 36.8 and 31.6% of the participants and for 100% of the participants during driving. The Letscom incorrectly recorded steps during deskwork but for a smaller proportion of the participants and for fewer participants than the Fitbit Alta did. However, the Letscom showed the largest percentage error, biggest range of error, and 95% of the participants had an error ≥25%. The Letscom also showed particularly poor agreement with manual step-count in terms of the limits of agreement. The limits of agreement were only wider for the Fitbit Alta. The large variability in error for individuals may explain why some participants commented on the Letscom being particularly inaccurate.

In agreement with previous findings that the objective numeric feedback provided by these devices can be a powerful motivational tool for some people with MS (18), the role of the devices as motivators was voiced in this study. This supports the potential use of wearable devices in physical activity behavior change interventions. However, the results of this study suggest that these commercially available devices may not provide valid estimates of step-count for adults with MS. Perceived inaccuracies of devices cast doubt on the value of them as monitoring tools and lead to frustration, distrust, and negatively affect people's motivation to use them (18).

Physical inactivity is a major global health concern for people with MS (24), with many people with MS spending two times as long seated compared to the general population (25). There is also a suggestion that improved physical activity levels among people with MS are indirectly associated with improvement in quality of life (26). As a result, key clinical guidelines specifically recommend tackling a lack of physical activity through targeted behavior change interventions (27). Activity monitors and pedometers are often used as an adjunct for such interventions and have been shown to increase motivation in home-based programmes, promoting increases in physical activity (18). However, it is pertinent that the validity and the acceptability of such devices are determined to aid device selection, especially for use among people with MS for whom 85% report gait deficits as their main presenting complaint (3). Although this study was conducted in a controlled environment, the tasks were chosen to capture common activities of daily living, including outdoor walking and driving where possible. This helps with the transferability of results beyond the laboratory environment into real-life scenarios, which has not been examined before. Considering that many people with MS use mobility aids and experience gait deficits (12), examining validity during simulated real-life scenarios, including navigating obstacles, is particularly important. However, future research should examine the validity of commercially available devices in free-living settings. A challenge when evaluating such devices is that technology is rapidly developing, with many devices being discontinued and replaced by new devices in short periods of time. Although the Fitbit Zip is the most commonly assessed commercially available device (11), it has been discontinued since this study was conducted. This somewhat limits the findings of this study. However, the findings from this study should be used as overarching principles to consider when developing or updating devices. The findings are also limited to ambulatory adults with MS, and, therefore, any recommendations for improving the validity and acceptability of these devices are not inclusive of wheelchair users.

CONCLUSION

In conclusion, commercially available devices demonstrated poor criterion validity when measuring step-count and time in activity among people with MS. This negatively affected the acceptability of devices, with perceived inaccuracies causing distrust and frustration. However, perceptions of how accurate devices varied between individuals, reflecting the large amount of variability in individual error observed during validation. Perceived accuracy was the overriding integral element to acceptability of these devices among people with MS. However, additional considerations when designing devices for people with MS include an appropriately sized and lit display and ease of attaching and charging devices. These considerations would potentially improve the acceptability and inclusivity of devices for all and not just people with MS.

DATA AVAILABILITY STATEMENT

The datasets presented in this article are not readily available because consent was not obtained from participants to share data. Requests to access the datasets should be directed to meriel.norris@brunel.ac.uk.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Ethical approval was provided by Brunel University London's Research Ethics Committee (REC) and the Health Research Authority and Health and Care Research West Scotland (REC reference 18/WS/0161). The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

GL, JFl, and JH completed data collection. JB and HJ contributed to data acquisition. JR, MN, and GL completed analysis and drafted the manuscript. JR, MN, GL, JFo, AS, and CK contributed to conception and design of the work. All authors contributed to the article and approved the submitted version.

FUNDING

This study was supported by funding from RCSI University of Medicine and Health Sciences STAR Programme and Brunel University London.

ACKNOWLEDGMENTS

The Fitbit Alta and Fitbit Zip devices used in this study were donated by Fitbit for the purpose of this study. Fitbit had no other involvement in the study.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fresc.2021.737384/full#supplementary-material

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Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers
in Rehabilitation Sciences

ORIGINAL RESEARCH
published: 23 March 2022
doi: 10.3389/fresc.2022.836655

Mobility Disability and Exercise: Health Outcomes of an Accessible Community-Based Center

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Objective: The purpose of this study was to determine how support and guidance provided by trained professionals during a 12-week, community-based transition exercise program, impact health outcomes and continued engagement in physical activity for persons with a mobility disability (PwMD).

Design: A single arm pre-post design was used.

Setting: Accessible community-based health and wellness center.

Participants: The study included 244 PwMD using a mobility device.

Interventions: Participants completed a 12-week transition exercise program provided through an accessible community facility that provided education and support to complete endurance and strength related exercises as well as programming to encourage transition to self-directed engagement in exercise.

Main Outcome Measures: Bodyweight, BMI, pain, perceived exertion, speed, and distance during cardiovascular fitness testing, and strength were measured pre and post exercise program. The number of participants that signed up for a monthly membership after the program was also monitored.

Results: For the total group, average pain reported over previous 30 days decreased significantly ($p < 0.01$), current daily pain decreased significantly ($p < 0.05$), perceived exertion at the end of the 9-min endurance test decreased significantly ($p < 0.05$), and the four upper extremity strength exercises showed large, significant strength gains ($p < 0.01$) after the program. There was no significant change in bodyweight, BMI, or speed and distance completed during endurance testing. At the completion of the program, 76% of participants enrolled in a monthly membership at the facility with the intentions to continue to exercise regularly.

Conclusions: This study provides evidence that an accessible community-based exercise program, with a transitional component supported by trained professionals, can support the exercise goals of PwMD and improve strength, decrease pain, and may promote regular exercise adoption for PwMD.

Keywords: exercise, mobility disability, strength, endurance, community-based research

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Specialty section:
This article was submitted to Disability, Rehabilitation, and Inclusion, a section of the journal Frontiers in Rehabilitation Sciences

Received: *15 December 2021*
Accepted: *25 February 2022*
Published: *23 March 2022*

Citation:
Morgan KA, Taylor KL, Walker CW, Tucker S, Dashner JL and Hollingsworth H (2022) Mobility Disability and Exercise: Health Outcomes of an Accessible Community-Based Center. Front. Rehabilit. Sci. 3:836655. doi: 10.3389/fresc.2022.836655

INTRODUCTION

According to data from the National Health Interview Survey, 5.8% of Americans aged 18–64 years have a mobility disability (1). Persons with a mobility disability (PwMD) are at a greater risk for major health conditions including cardiovascular disease, hypertension, and diabetes compared to those without disabilities (2–4). One of the major contributors to these health disparities experienced by PwMD is that they are more vulnerable to secondary conditions that result in an increased number of hospitalizations and high costs for treatments that could be prevented through improved levels of exercise. The number of PwMD will continue to grow, as the number of people using wheelchairs is predicted to quadruple between 2005 and 2030 (5). Compared to other disability groups (vision, hearing, and cognition), adults with a mobility disability have the highest prevalence of inactivity, with over half reporting inactivity (1).

Regular exercise is widely recognized as having health benefits (2, 6–8) and is also connected with more established social networks, greater participation in life activities, and greater likelihood of employment (9–11). However, PwMD remain one of the least physically active populations in the U.S., and those who are active, often are not experiencing the health-related benefits of exercise (12, 13). The typical daily routine of PwMD does not produce positive health-related changes such as cardiovascular increases; therefore, structured exercise activities are needed to promote health-related benefits (14). Consistent participation in exercise is a difficult area of reintegration for PwMD outside of the traditional clinical setting (15), with decline in functional capacity often occurring after discharge from rehabilitation (9). A gap in the continuum of care exists from rehabilitation to the community; therefore, PwMD often lack the appropriate guidance and resources to achieve successful exercise goals following completion of therapy (10).

PwMD commonly experience physical or program barriers that limit or prevent them from accessing health and wellness programs outside of the medical and rehabilitation model (10, 16). The barriers to participating in regular, structured exercise, outside the medical and rehabilitation setting, are well documented for PwMD. Environmental barriers to exercise for PwMD include considerable lack of accessible facilities, equipment, supports, and lack of trained, knowledgeable staff (10, 15, 17–22). Personal barriers to exercise participation include lack of information on available exercise programs and accessible facilities in the community; lack of experience with exercise equipment, programming and techniques; and reduced motivation to voluntarily participate in physical exertion. PwMD have also reported being inundated regarding initiating an exercise program, especially if they were not familiar with exercise techniques prior to their disability (17, 18, 23, 24). PwMD may also experience a perceived conflict with the cultural norms of traditional community gyms, leading to negative interactions with staff and other gym participants (23).

Traditional home exercise programs, often prescribed at discharge from rehabilitation, may overcome initial participation barriers but often do not provide support for initiating an exercise program designed to promote health-related benefits, supervision during physical activity, ongoing education or an optimally individualized physical activity prescription, which improves outcomes, progression, and safety monitoring (25). Researchers continue to explore methods and strategies to engage PwMD in physical activity, and common shortfalls of these approaches include a lack of professional support, poor adherence rates, inability to maintain physical activity increases, and tested interventions not translating into sustainable models (25, 26). Thus, transition from clinical evidence of exercise effectiveness to provision and establishment of effective community-based exercise programs (CBEPs) for PwMD has proven challenging. More evidence is needed to identify evidence-based exercise approaches implemented in the community that improve the participation of exercise for underserved PwMD (15).

To address barriers and promote physical activity participation among community-dwelling PwMD, CBEPs need an adequate combination of participant education; individualized programming based on evidence-based recommendations; knowledgeable, trained support staff (11); and accessible equipment within an accessible facility. CBEPs may be an essential component in the continuum of care to monitor and optimize health, function, and participation for PwMD (10). Furthermore, it is worthwhile to determine how to best support PwMD to establish routines as life-long exercisers, prevent secondary health conditions, and promote their overall well-being. Therefore, the purposes of our study were to determine (1) the prevalence of participants who remained engaged in regular exercise following completion of a 12-week, community-based, transition exercise program and (2) health-related outcomes of participating in the 12-week CBEP for PwMD. Our study seeks to fill a gap in the literature regarding health outcomes and exercise engagement for PwMD successfully participating in supportive, accessible CBEP.

MATERIALS AND METHODS

Study Design

A pre-post, single arm, prospective, within-subject design was used.

Setting

The long-running research study took place from March 2006 to October 2017 at an accessible exercise facility in the Midwest region of the United States operated by a disability organization. The accessible exercise facility component of the organization was staffed by trained healthcare professionals including occupational therapists, occupational therapy assistants, and a physical therapy assistant. The organization also has relationships with several local higher education institutions, from which the exercise facility accepts health professional graduate students

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Abbreviations: 1-RM, 1-repetition maximum; ArmE, Arm/Leg Ergometer; CBEP, Community-based exercise program; CORE, Characteristics of Respondents survey; ICF, International Classification of Functioning, Disability and Health; PwMD, Persons with a mobility disability; RPE, Rating of perceived exertion; SCI, Spinal cord injury; T-1, Baseline assessment; T-2, Terminal assessment; VO_{2peak} Peak oxygen consumption; W, watts.

for clinical fieldwork rotations, regular community service engagement, and other capacities.

Participants

Potential participants were recruited *via* flyers, advertisements, referrals from local rehabilitation facilities, and word-of-mouth. Participants were eligible for inclusion if they: (1) were 18 years or older, (2) had a mobility disability requiring use of a mobility device, (3) were community-dwelling, and (4) were able to provide informed consent. The International Classification of Functioning, Disability and Health (ICF) model was used to frame the basis for participant eligibility related to mobility disability instead of medical diagnosis (27). The following ICF codes were used as inclusion criteria to recruit and enroll a non-representative convenience sample of PwMD: b730 Muscle power function, d450 Walking exclusion, d465 Moving around using equipment, and e120 Use assistive mobility device (28). The project was approved by our university institutional review board. Persons were excluded from the study if they did not have a medical condition requiring use of a mobility device (cane/crutch(s)/walker, manual wheelchair, power wheelchair, scooter); were under 18 years of age, lived in a facility such as a nursing home, were unable to provide informed consent, or unable to provide a physician release to exercise.

Community-Based Exercise Intervention Program

The 12-week exercise program consisted of 1:1 or 2:1 guided and supervised exercise training by trained staff. Staff included occupational therapists, an occupational therapy assistant, a physical therapy assistant, and graduate students from healthcare programs. The exercise program was based on the American College of Sports Medicine's Physical Activity Recommendations (29, 30). The intervention included 1–2 h sessions, one-to-three times per week, for a goal of 12 weeks. Prior to each exercise session, participants' current pain level was recorded. While the 12-week program was centered on each participant's goals, abilities, and preferences, exercise sessions maintained a foundational structure. Each exercise session included a warm-up, opportunities to do cardiovascular and strength exercises and a cool-down. Many participants also chose to do flexibility training during their sessions through range-of-motion and stretching exercises.

The primary goal of the program was for participants to self-direct their own exercise regimens following the 12-week program. The exercise program followed three adaptable phases that progressed at varying rates depending on individual participants: (1) education and setup, (2) guidance and assistance, and (3) transition and monitoring. During the education and setup phase, staff educated participants on physical activity recommendations and various exercise modes, and provided instruction and demonstration on proper equipment setup and exercise technique. The majority of the exercise program consisted of the guidance and assistance phase. During this phase, staff provided verbal, visual, and/or physical support to assist participants during their exercise sessions including transfers, equipment setup, spotting, or adjusting exercise techniques. The transition and monitoring phase occurred throughout the program but became the focus during the final 2–3 weeks. Staff provided less guidance during exercise sessions, promoting participants' autonomy and self-monitoring. For example, participants might prefer more cardiovascular workouts to achieve their goals and choose to complete both the Vitaglide and the arm ergometer while only completing a few of the strengthening exercises (biceps, rickshaw).

To ensure fidelity and consistency, study protocol and procedures were maintained throughout the study. During the time period of this study, changes included addition of new equipment and a few changes in staffing. Quarterly staff trainings were conducted to ensure that all staff were consistent with testing and intervention protocol and procedures. To maintain data collection fidelity, testing and workout tracking forms were employed to guide staff on protocol delivery and data documentation.

Exercise Equipment

Participants used a variety of equipment during the 12-week exercise program. Strength equipment included the Uppertone (GPK Inc., El Cajon, CA, USA), Equalizer (Equalizer Exercise Machines, Red Deer, Alberta, CA), free weights, and resistance bands. Endurance equipment included the Endorphin Arm Ergometer (ArmE; Pro-Med Products, Alpharetta, GA, USA), Motomed (RECK-Technik GmbH & Co., Betzenweiler, DE), Vitaglide (Planet Mobility, Shelby Township, MI, USA), manual wheelchair rollers (provided by Dr. Rory A. Cooper, PhD, University of Pittsburgh, Pittsburgh, PA, USA), standard treadmill (Planet Mobility, Shelby Township, MI), and NuStep (NuStep, LLC, Ann Arbor, MI, USA). All exercise equipment used for assessments and exercise sessions was accessible for PwMD. During assessments, participants were tested to measure their baseline strength using either the Uppertone or Equalizer, and tested to measure their baseline endurance using the ArmE. During exercise sessions, participants had access to any of the available equipment in the facility. Descriptions of the equipment can be found in **Appendix A**.

Procedures

Eligible participants attended a workshop, where they were screened for eligibility, given information on the facility and the 12-week exercise program, toured the facility, and provided informed consent. Enrolled participants were required to obtain physician's release prior to beginning exercise.

Testing Protocol

All study participants completed baseline (T-1) testing prior to and terminal (T-2) testing at the completion of the 12-week program. Bodyweight, resting vitals, and current and average pain were measured using assessment equipment and established outcome measures below.

The cardiovascular endurance test was performed with the upper extremities on the ArmE. Participants transferred to a seat or sat in their mobility devices to perform the test. For participants who were unable to grip the ArmE handles, grip assists, Ace bandage wraps, or neoprene gloves were used to secure their hands. The asynchronous ArmE protocol included an initial 30-s speed test to establish a testing speed the participant perceived as "hard," or 5/10 on the Modified Borg RPE scale (31). The speed test was followed by the 9-min graded exercise test, in which participants were instructed to maintain the established "hard" speed throughout the test. All participants initiated the test at 19 W, with incremental increases by 4 W every 3 min for 9 min. Participants were asked to rate their perceived exertion (1–10) (32) at the end of minutes three, six, and nine and following completion of the test.

Strength testing consisted of establishing a 1-repeition-maximum (1-RM) on four upper extremity exercises (chest press, back row, biceps curl, and rickshaw triceps extension) performed unilaterally. The highest amount of weight pushed or pulled through a complete range of motion was recorded as the 1-RM using the Uppertone, or Equalizer.

Outcome Measures

Cardiorespiratory fitness was assessed using a 9-min, incremental test with the ArmE by measuring average speed at minutes three and nine (m/s), self-reported perceived exertion of the 3rd and 9th min (RPE; 1–10) (32) and total distance completed (m). Strength was assessed *via* unilateral 1-RM (kg) across four upper extremity exercises (chest press, back row, biceps curl, and rickshaw) unilaterally with the right upper extremity, using either the Uppertone or Equalizer. Weight was measured using a Seca model 664 digital wheelchair scale. The average level of pain for the previous 30 days was assessed at each assessment using the Wong-Baker FACES Pain Rating Scale (0–10) (33, 34). The Characteristics of Respondents survey (CORE) (35) was also used to gather demographic information (**Table 1**). Interest in continuing to exercise was measured by whether or not the participant signed up for a monthly membership to the facility following completion of the program.

Data Analysis

Statistical analyses were conducted using IBM SPSS Statistics (Version 26). Demographic information was compiled using descriptive statistical methods. Paired sample t-tests were used to compare outcome measures (bodyweight, current pain, average pain over 30 days, cardiovascular fitness, and strength) between baseline and terminal assessments. Dependent measures for cardiorespiratory fitness included average speed and RPE during the 3rd and 9th min and total distance completed. Equipment used to perform the strength testing protocol changed from the Uppertone to the Equalizer in October 2009. Therefore, strength data were separated and compared according to equipment used. Pre-post 1-RM for each of the four upper extremity exercises using the right arm, were used as dependent measures to determine changes in strength. Statistical comparisons were conducted for the total group, as well as between session frequency intensities (intermittent v. concentrated). *Intermittent* participants completed an average of one session per week, while *concentrated* participants completed an average of two to three sessions per week. Outcome measures were also compared among the three most frequent diagnoses: spinal cord injury, stroke, and multiple sclerosis. Due to mitigating factors impacting many participants' ability to complete 2–3 sessions per week, program duration inclusion was expanded to include participants who completed the CBEP in 12–18 weeks with a minimum of ten sessions and maximum of 36 sessions. Values are expressed as mean ± SD, unless otherwise stated. Two-tailed significance was accepted at $p < 0.05$.

RESULTS

Over the course of the long-running CBEP study, 348 participants completed the 12-week program and both testing

TABLE 1 | Assessments made prior to (T-1) and after T-2) exercise intervention.

Measure	Instrument/device	Unit of measure
Demographics	CORE survey	
Fitness		
Body Weight	Seca model 664 wheelchair scale	pounds (converted to kg)
Body mass index (BMI)	Calculated	weight/height
Current pain level	Faces Pain Rating Scale	1–10 (high)
Average pain level over past 30 days	CORE	1–4 (high)
Endurance		
Speed of arm crank turn (ArmE)–3 min	Endorphin® Arm/Leg Ergometer	meters/second
Speed of arm crank turn (ArmE)–9 min	Endorphin® Arm/Leg Ergometer	meters/second
Rate of perceived exertion (RPE)–3 min	Modified Borg Scale	1 to 10 (high)
Rate of perceived exertion (RPE)–9 min	Modified Borg Scale	1 to 10 (high)
Strength/resistance		
Biceps	Uppertone or Equalizer	repetition maximum (1-RM)
Chest press	Uppertone or Equalizer	repetition maximum (1-RM)
Rickshaw triceps extension	Uppertone or Equalizer	repetition maximum (1-RM)
Rowing left	Uppertone or Equalizer	repetition maximum (1-RM)

ArmE, arm ergometer.

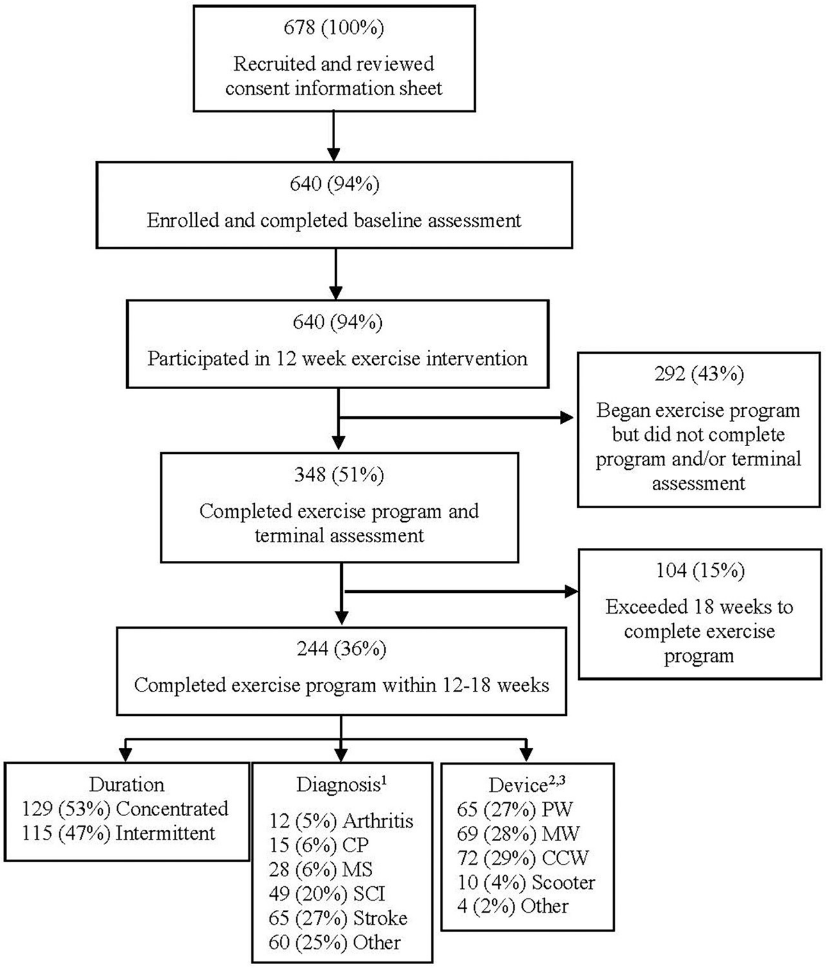

FIGURE 1 | Participant recruitment, enrollment, and study participation flow diagram. [1]Refused to answer the diagnosis survey question ($n = 15$, 11%). [2]PW—power wheelchair, MW—manual wheelchair, CCW—cane, crutches, and walkers. [3]Refused to answer the device-used survey question ($n = 24$, 10%).

sessions and 244 of these participants met the eligibility criteria for analysis (**Figure 1**). Two hundred forty-four participants completed both assessments and finished the 12-week exercise program within 12–18 weeks (**Table 2**). There was equal representation of gender and nearly equal of race (48.8% White and 41.8% non-white) with the majority being low income and fairly high levels of education. A mean 20.4 ± 5.5 (range 10–34) total sessions were completed over an average 13.5 ± 1.8 weeks. Weekly frequency of exercise sessions varied from one to three, with an average 1.54 ± 0.5 sessions per week. Out of 244 participants, 76% ($n = 186$) expressed interest in continuing to exercise by enrolling in the monthly membership program at the facility.

Total group results ($n = 244$; **Table 3**) of the 12-week exercise program showed no significant changes in bodyweight or BMI. Current pain and 30-day average pain decreased significantly ($p < 0.05$ and $p < 0.01$, respectively). A statistically significant decrease in RPE was achieved at minute-nine of the ArmE endurance test ($p < 0.05$). Total distance completed on ArmE endurance test increased by 11%; however, this was not significant. Strength significantly increased across all four strength exercises regardless of equipment used. Increases in strength for all exercises exceeded 10%. No significant differences were found between workout frequency intensities or among diagnoses.

DISCUSSION

Although exercise intervention research for people with disabilities has become more prevalent in the last decade [increasing by 60% since 2010; (36)], most studies are diagnosis-specific, significantly limiting sample size and reducing generalizability to the overarching population of PwMD. Most CBEPs are also limited by often targeting ambulatory populations, lack of customizable programming for neurological diagnoses other than stroke, and little structure for individuals to continue exercising after the formal intervention period is

TABLE 2 | Demography of intervention sample ($N = 244$).

	Mean (SD)	Range
Age (241 responded)	51.12 (15.41)	17-88
Gender	***n***	**%**
Female	121	49.6
Male	121	49.6
No answer	2	0.8
Race		
Black/African American	102	41.8
White	119	48.8
Other	14	5.7
No answer	3	1.2
Personal annual income		
\$0–\$14,999	101	41.4
\$15,000–\$34,999	62	25.4
\$35,000–\$54,999	17	7.0
\$55,000 or more	16	6.5
No answer	48	19.7
Highest grade completed		
Grade 1–11	23	9.4
Grade 12/GED	59	24.2
College 1–3 years	75	30.7
College ≥ 4 years	80	32.8
No answer	7	2.8
Primary disability		
Arthritis	12	4.9
Cerebral palsy	15	6.1
Multiple sclerosis—MS	28	11.5
Spinal cord injury—SCI	49	20.1
Stroke	65	26.6
Other conditions	60	24.5
No answer	15	6.1
Device		
Power wheelchair	65	26.6
Manual wheelchair	69	28.3
Cane/crutches/walker	72	29.4
Scooter	10	4.1
Other devices	4	1.6
No answer	24	9.8

SD, standard deviation; GED, general education diploma.

complete (9). The current study sought to provide a staff-guided, multi-modal transitional exercise program in an accessible, community-based facility and assess effectiveness in improving health-related outcomes and to promote adoption of a physically active lifestyle among PwMD who often cannot successfully utilize traditional fitness centers and gyms.

Providing direct professional support and guidance alone may not directly result in long-term behavior change. Interventions rooted in self-determination theory (SDT), which focus on cultivating autonomous motivation, a sense of belongingness, and confidence in one's actions, have shown promise in promoting physical activity—related behavior change (37, 38). Similarly, behavior change techniques (BCT), or systematic intervention methods used to change psychological determinants of behavior (e.g., self-efficacy, health beliefs), are commonly used in interventions for physical activity for spinal cord injury research. The theoretical mechanism of action for self-management is self-efficacy, which was promoted in this study *via* the three-phase transitional CBEP, particularly the transition and monitoring phase, may have contributed to a high percentage of participants wanting to continue to exercise (39).

Evaluation of behavior change is becoming more prevalent related to healthy lifestyle adoption among PwMD (36). Interventions that support autonomy and self-efficacy in exercise participation have been shown to promote self-management and increase the probability of implementing physically active behaviors, independently (40). The current study integrated a transition and monitoring phase to support participants' autonomy and ownership over their exercise routines to promote continuation of regular physical activity participation. Seventy-six percent of participants successfully transitioned and maintained their exercise regimens following completion of the 12-week program. The transitional component of an exercise program is critically important to lifelong engagement in physical activity for PwMD (9). The adoption of an ongoing physical activity regime has been shown to decrease secondary conditions and improve overall health, thus making the investigation of the successes found in this CBEP important to analyze in order to determine how to replicate widely.

Large, significant increases in strength occurred across all four exercises, which align with previous research (41–43) and further support the musculoskeletal benefits of consistent participation in structured, individualized strength training in CBEPs for PwMD. Participants demonstrated a mean increase of over 5.9 kg (23%) in strength for back row and rickshaw triceps extension exercises. Improved performance on these two exercises is particularly important for individuals using wheeled mobility devices, as they counterbalance muscles frequently used for wheelchair propulsion, as well as strengthen muscles used during functional transfers (44).

Minimal changes in cardiorespiratory fitness occurred during the study, with a decrease in RPE for minutes-three of the ArmE endurance test being the only significant result. These results differ from previous clinical studies of endurance exercise interventions, which have shown improved cardiorespiratory fitness (36, 42, 43, 45). Following recommended guidelines (30), measurable increases in cardiovascular health can occur within 8–12 weeks (46–49); however, a minimum exercise intensity threshold must be met. Exercise intensity is a key component for changes in aerobic capacity, with moderate-to-vigorous intensity being the recommended threshold for the frequency and duration of the current study (30). Intensity was only measured during assessments and was not regulated during exercise sessions.

Minimal change in cardiorespiratory fitness may also be explained by methodological limitations. Peak oxygen consumption (VO_{2peak}) during a graded exercise test is currently the gold-standard for assessing cardiorespiratory fitness and is often used in studies examining endurance changes in PwMD (50, 51). VO_{2peak} testing assesses changes at the metabolic level,

TABLE 3 | Influence of exercise on total intervention group (*N* = 244).

Variables	*n*	T-1 Mean (SD)	T-2 Mean (SD)	T-2-T-1	% Δ
Body weight (kg)	134	85.0 (28.6)	85.8 (27.5)	0.8	0.9
BMI	130	29.5 (9.3)	29.8 (9.0)	0.3	1.0
Current pain	230	2.1 (2.6)	1.7 (2.3)	−0.4	−23.5*
Average pain	223	3.2 (2.7)	2.3 (2.9)	−0.9	−39.1†
Arm ergometer (ArmE)					
ArmE speed: 3 min (m/s)	214	7.0 (4.1)	7.2 (3.2)	0.2	2.7
ArmE speed: 9 min (m/s)	203	5.7 (3.9)	5.7 (3.0)	0.0	0.0
ArmE RPE: 3 min	232	3.2 (1.7)	3.2 (1.7)	0.0	0.0
ArmE RPE: 9 min	219	5.6 (2.1)	5.3 (2.1)	−0.3	−5.7*
ArmE total distance (km)	211	1.6 (1.0)	1.8 (3.2)	0.2	11.1
Uppertone (*n* = 96)					
Biceps (kg)	81	13.7 (9.1)	15.3 (9.3)	1.6	10.4†
Chest (kg)	88	22.8 (11.2)	26.8 (10.4)	4.0	14.9†
Rickshaw (kg)	86	16.6 (8.2)	20.9 (9.5)	4.3	20.6†
Rowing (kg)	90	23.0 (10.2)	28.0 (9.6)	5.0	17.9†
Equalizer (*n* = 148)					
Biceps (kg)	62	10.7 (6.8)	13.0 (7.1)	2.3	17.7†
Chest (kg)	76	15.5 (10.2)	21.0 (11.9)	5.5	26.2†
Rickshaw (kg)	128	23.0 (13.4)	30.5 (16.7)	7.5	24.6†
Rowing (kg)	76	20.0 (9.7)	26.0 (12.3)	6.0	23.1†

SD, standard deviation; ArmE, arm ergometer; min, minutes; m/s, meters per second; kg, kilograms; km, kilometers, $^{*}p < 0.05$, $^{\dagger}p < 0.01$.

providing greater sensitivity to changes compared to the ArmE endurance testing protocol administered in this study. Previous studies that used similarly broad endurance testing methods reported minimal-to-no significant changes in cardiorespiratory endurance (9, 11). CBEPs typically do not have access to the equipment and advanced training required to reliably conduct metabolic testing. Despite the lack of change in cardiorespiratory fitness, the significant decrease in RPE indicates reduction of perceived effort at the same exercise intensity, which may be an indicator of improved endurance. The CBEP, described in the current study, promoted self-efficacy among participants with the objective of improving self-directed participation in an exercise program. Previous evidence supports the relationship between self-efficacy and improved changes in RPE (52).

Varied dosing-frequency may have also contributed to limited changes in cardiorespiratory fitness. While the curvature of the dose-response relationship remains ambiguous, numerous studies have concluded the health benefits of regular physical activity (53) directed by pre-established guidelines (4, 25). Similar to exercise intensity, a minimum frequency threshold must be met to elicit measurable improvements in cardiorespiratory fitness. Intermittent participants (average of one session/week) comprised 47% of our included sample; as the recommended frequency of moderate-vigorous cardiovascular exercise is 3–5 days per week (4, 25), these intermittent participants likely did not meet the minimum threshold to produce measurable improvements in cardiorespiratory health.

No significant changes in weight were noted, similar to some previous studies (11, 54–56) and different from others (47). Previous studies found similar results related to BMI (54, 57). Pain (both current and average over previous 30 days) decreased significantly. Limited evidence exists examining the effects of exercise on pain reduction for PwMD; however, available literature supports the present study's findings in acute and chronic pain reductions post-exercise intervention (58–61).

Study Limitations and Future Directions

The current study included limitations in methodological rigor and outcome measures. This study lacked a control group limiting the causative inferences of the CBEP. The lack of changes in cardiorespiratory fitness may reflect the inadequate sensitivity of the measures used. While this is a limiting factor of the study, practicality should be considered for community-based settings; future disability and physical activity research should explore alternative methods for measuring fitness that are reliable and sensitive but also practical in a community-based setting, as this aligns with current National Institute of Health recommendations (62).

One of the primary objectives of this study was to determine the prevalence of participants who remained engaged in regular exercise after completing the CBEP, which yielded a 76% success rate. The current study did not include a formal outcome measure of self-efficacy to attempt at measuring continued success outside of the CBEP such as quantitative questionnaires or qualitative interviews. Future studies should utilize such assessment tools to evaluate self-efficacy and autonomy at baseline and at distinct timepoints throughout participation in exercise to future guide development of CBEPs for PwMD.

Self-direction is a fundamental difference between clinic-based exercise programs and CBEPs. CBEPs, like the present study, allow participants to select exercise equipment based on their personal goals and preferences, resulting in increased variability of equipment used and exercises performed. For example, endurance exercises had no specific time, distance, or intensity per session and were variable by participant. In contrast, for strength exercises, support staff often set a distinct number of sets and repetitions at specified weights for participants. A dose-response analysis was conducted based on weekly frequency, but no significant differences were found, likely confounded by the variability of exercises completed across participants. Weekly frequency was also often dependent on several variables including transportation availability, health status, personal assistance and work schedules, and support staff availability. Time to complete the 12-week program also varied to accommodate for mitigating factors impacting many participants' ability to complete two to three exercise sessions every week. Due to the importance of individualization of exercise programming for PwMD, future studies should incorporate reliable and sensitive methods for tracking intensity during testing and intervention to further customize participants' exercise frequency and duration based on intensity achieved. Future studies should also consider tracking participants' daily activity outside of the exercise sessions to examine any changes or differences in activity patterns in PwMD's daily lives.

CONCLUSIONS

Exercise is one of the vehicles for community reintegration and participation for PwMD; however, physical inactivity remains one of the hallmark traits of this population (63). This study provides evidence that an individualized CBEP can significantly improve upper extremity strength and decrease pain for PwMD, as well as effectively transition PwMD from a formally guided program to a self-monitoring continuation of regular physical activity. The program in this study provided knowledgeable, professional support; customized programming; and an accessible facility and equipment, which are integral for PwMD to participate in CBEPs. Lack of change in cardiorespiratory fitness was likely attributed to methodological limitations of the endurance test, inadequate achievement of minimally-recommended exercise intensity and frequency, and decreased sensitivity of the methods used to assess fitness and monitor intervention intensity. PwMD require access to accessible, community-based fitness programs post-rehabilitation to continue recovery, reduce risk of comorbidities and mortality, and optimize functional independence, societal participation, and overall quality of life.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/**Supplementary Material**, further inquiries can be directed to the corresponding author/s.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Washington University Institutional Review Board. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

KM assisted with original conception and design of the study and assisted with data analysis and interpretation of the data. She drafted the article and provided final approval to the draft being submitted for publication. KT was involved with analyzing and interpreting the data. She provided intellectual content related to all sections of the paper and provided final approval to the draft being submitted for publication. CW was involved with providing technological support to the acquisition of data. She contributed intellectual content to the methods, results and discussion sections and provided final approval to the draft being submitted for publication. ST and JD were involved with data collection. They contributed intellectual content to the methods section and provided final approval to the draft being submitted for publication. HH was involved with designing the study, data analysis and interpretation of the data. He contributed intellectual content to the methods and results sections and provided final approval to the draft being submitted for publication. All authors contributed to the article and approved the submitted version.

FUNDING

Missouri Foundation for Health (04-0551); US Department of Education, NIDRR (H133A010701); and National Institutes of Health, Comprehensive Opportunities in Rehabilitation Research Training (K12 HD055931).

ACKNOWLEDGMENTS

This work was developed and initiated by the vision of the late disability researcher, David B. Gray, PhD. We would like to thank Kim Walker, Denise Dickerson, Mike Scheller, Lindsey Kampwerth, Melissa Smith, Megen Devine, and the many occupational therapy students and Paraquad employees who assisted in various aspects of the research. Finally, we wish to extend a deep expression of gratitude to all of our participants.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fresc.2022.836655/full#supplementary-material

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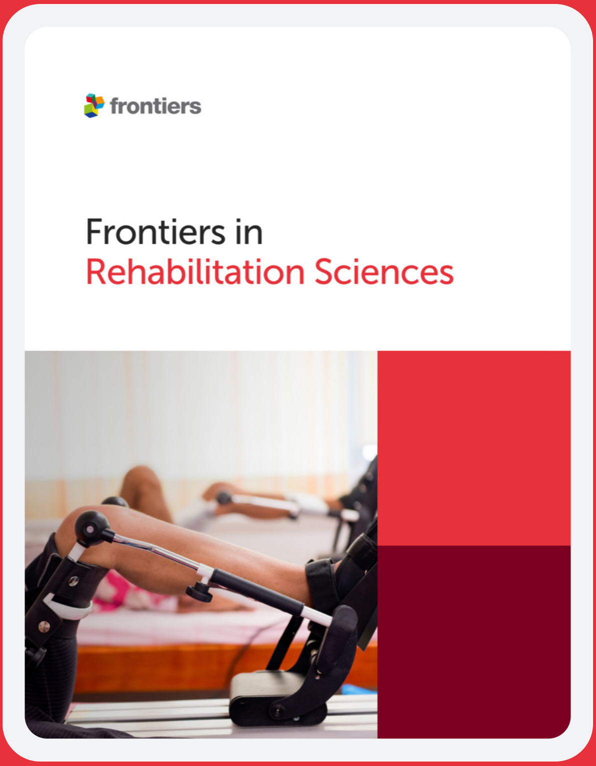
Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.



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