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An exploration of physical activity experiences throughout the Huntington's disease journey: supporting development of theoretically underpinned complex interventions

Physical activity in Huntington's disease

Research Paper

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An exploration of physical activity experiences throughout the Huntington's disease journey: supporting development of theoretically underpinned complex interventions

Abstract

Purpose. Huntington's disease is an autosomal dominant neurodegenerative disease. Progressive physical, behavioural and cognitive impairments cause loss of independent function. Physical activity interventions are important components of comprehensive intervention strategies and may help alter the functional decline trajectory. Qualitative research has an important role to play in developing theoretically sound, well-defined physical activity interventions in Huntington's disease.

Materials and methods. Eight focus groups were conducted with people with prodromal to late stage Huntington's disease, caregivers (family members/formal), and healthcare professionals. An analytical coding framework was developed from the data and Levanthal's self-regulation model to assist analysis.

Results and Conclusions. Key themes were identified: evolving representations of Huntington's disease and physical activity; varying social environment of the person with Huntington's disease and the impact on physical activity; achieving physical activity participation while coping with the nuances of Huntington's disease. Levanthal's model facilitated understanding of physical activity experiences, however with progression, self-regulation of activities needs to become more collaborative with caregivers. A modified self-regulation model specific to physical activity in Huntington's disease is presented.

Using a novel approach to generate new understanding of physical activity across the Huntington's disease lifespan facilitated development of an original and significant theoretical foundation to underpin development of a range of much needed physical activity and exercise interventions in Huntington's disease.

Key words

Huntington's disease, physical activity, exercise, experiences, complex interventions, qualitative, theoretical framework, illness perception, Self-Regulation Model

Introduction and Background

Huntington's disease (HD) is a devastating neurodegenerative disease. Progressive physical, behavioural and cognitive impairments eventually lead to inability to function independently. Death usually occurs death 15-20 years post symptom onset (1–3). Physical activity and exercise interventions are vital for maintaining function and independence, and specific to HD, evidence supports physical activity interventions for benefit to quality of life and fitness (4–7). It is important to distinguish between the terms 'physical activity and 'exercise' as they are often used interchangeably. Exercise is a subcategory of physical activity and is planned, structured, repetitive and purposeful with the goal of maintaining or improving components of physical fitness. Physical activity comprises not only exercise but other recreational activities, work, household chores, play, and active transport that involve bodily movements [57]. For the purposes of this research, the term physical activity was used when discussing physical activity and exercise.

Physical activity is a complex intervention comprising many interacting components that influence uptake and maintenance. In HD, symptoms in terms of behaviour, cognition and physical ability further compound the complexity of delivering such interventions. Previous studies of physical activity and specific exercise interventions in HD have not considered in detail how these factors affect adherence and outcome despite the fact that tailored interventions are likely to be more effective (8). Such interventions need to account for context (e.g. pre-existing conditions or anything external to the intervention that may

affect the outcome such as beliefs, values, attitudes, previous experiences, support, or social /cultural norms) and complexities of HD as a disease.

Only a small number of published HD physical activity and exercise interventions have included qualitative research or process evaluations (4,5,9). These studies highlight relatively generic themes in line with the general physical activity literature in healthy populations. For example, themes such as 'facilitators and barriers', but there was little exploration of disease specific context. The methodological limitations of these studies mean that further in-depth qualitative research is needed.

Studies that have explored experiences of living with HD highlight complexities in daily life (10,11). The impact of physical, emotional, cognitive, behavioural and psychological impairments leads to feelings of loneliness, hopelessness, emotional avoidance (denial), stigma and guilt about passing on the gene. A focus group study with people with HD, family members, and carers found that reduced ability to communicate leads to social isolation and is a challenge for carrying out activities of daily living (12). Chorea was perceived to be distressing for others to see rather than being distressing and disabling to the person with HD. Carers highlighted this by describing use of wheelchairs for people with choreic gait because of fall concerns (10). HD has a cost to health and quality of life of caregivers due to the psychosocial consequences of the strain of daily, and long-term responsibility of care in HD, isolation, and stigma of HD (13–16). The complexities of these issues and experiences have implications for how people with HD engage and participate in physical activity which in and of itself is complex, and also for research into physical activity and specific exercise interventions.

Amongst the few studies that explore lived experience of HD, Levanthal's self-regulation model and illness perceptions (a component of the self-regulation model) have

been used (17–19). The self-regulation model depicts self-regulation as a processing system where individuals move through stages of information from when a health threat is received, representations of the threat are formed, acted upon (problem solving, goal planning, action plan), evaluated and then perceptions are integrated into memory. Vital components of the self-regulation model are illness representations, namely 'cause', 'control', 'identity', 'timeline' and 'consequences'. These representations develop from different sources of information, including an individual's personal experience of the illness, illness-related beliefs, and the social and cultural context. Information about illness is gained through public media; the news, social media, through face-to-face communication with family, friends and health care professionals. The illness representations of timeline and identity are particularly pertinent considering issues surrounding identity and changes with progression of HD, and the concept of coping has been elicited as a major part of life with HD (14,20,21). The self-regulation model (19) has helped in furthering understanding experiences related to psychosocial and physical consequences of HD (17,18,20–22) and could provide further understanding of physical activity experiences in HD and underpin tailored physical activity interventions.

Studies in HD using the self-regulation model found that people with HD have a strong illness identity, perceive themselves to have little control in a personal sense and in terms of treatment, and characterise HD as an illness of long duration. Beliefs about HD appear to play a role in well-being and so enhancing an individual's perception of control may be important to their quality of life (21). Positive coping mechanisms such as use of emotional and instrumental support and finding the positives in situations were identified (17,18).

In order to develop relevant, realistic and appropriately supportive physical activity interventions, it is vital to understand the prior personal experiences of people with HD. The

aim of this study was to explore how living with HD impacts on the experience of physical activity across the stages of the disease and to identify a theoretical framework that informs development of theoretically sound physical activity interventions for HD.

Methods

A qualitative approach was used to capture experiences of physical activity in HD. Focus groups were considered appropriate based on the research aim of obtaining broader experiences across the stages of HD and have previously been useful in exploring experiences of HD (10, 12, 23). In developing the research design, potential challenges of conducting research with people with HD were considered and are discussed in the following sections. Ethical approval to conduct the research was obtained from the School of Healthcare Sciences, Cardiff University.

Participants

Participants were recruited to eight focus groups through the Huntington's Disease (HD) Association of England and Wales. Specialist HD association advisors in each region were approached to help run focus groups with people with HD and their caregivers to discuss physical activity experiences.

Participants included people with prodromal to late stage HD, and caregivers (family members/formal). Caregivers were included because the impact of HD is experienced by people with HD and those close to them giving them much insight (24). Inviting people across the spectrum of HD meant that participants may become upset by seeing others at later stages, so on attending the focus group sessions, participants were allowed to naturally form groups by choosing which table they sat at. The natural forming of groups suggested that participants were comfortable with each other and had begun to develop interactions, which may have been more conducive to rich focus group discussions. This could facilitate a

dynamic that would attenuate ‘power differences’ between the researcher and participants (25). As such, the groups that had formed were not changed. Previous studies using focus groups grouped together family members and caregivers, people with HD at similar stage, and people across the stages of HD (10, 12, 23). Informed written consent was obtained from participants before the start of the focus groups.

Data collection

Discussions were guided by a schedule designed to explore experiences of physical activity and impact of HD on participation. Questions were developed following a literature review that identified key features of experiences of living with HD and physical activity experiences in HD and neurodegenerative conditions (9,20,21,26–32). Key features included aspects of HD progression, motivation, strategies for engagement, barriers and facilitators to physical activity, and illness identity. Following the pilot focus group, the first question was simplified. Focus groups lasted approximately 1 hour and were audio taped and transcribed verbatim (removing any identifiable data). Basic participant characteristics were collected to describe the focus groups (table 1). As recruitment was via the HD Association, some participants knew each other through attending HD Association branch meetings. Focus groups were conducted by research physiotherapists who work with people with HD and knew some of the participants in focus groups 1, 2 and 3. The local specialist Huntington’s disease advisors who had helped recruit participants were also present during the focus group sessions.

It was possible that during data collection not all voices would be heard equally, particularly because people with HD often have difficulty initiating conversation (33). Caregivers asked for permission to speak on behalf of some HD participants and make

clarifications. The caregiver presence, particularly for those with late stage HD could help ameliorate the possibility of memory and communication deficits affecting the patients' recall and expression of experiences (1). This approach has been used with stroke patients who may have had similar issues (34). In order to include people with late-stage HD this was something that had to be accepted. However, all caregiver statements were checked with the person with HD for either a verbal or non-verbal indication of agreement (35). Information related to function, time since onset of symptoms and independence was collected prior to the focus group session in a short questionnaire. The questions were framed around the total functional capacity scale (58) so that approximate disease stage of participants could be identified.

Insert <Table 1. Focus group demographics>

Data Analysis

Framework analysis adapted from Spencer and Ritchie (36) was used to develop key themes. Stages of framework analysis include familiarisation with the data, developing and refining an analytical framework, indexing data using the analytical framework, charting data into matrices, and mapping and describing. Memos were used throughout analysis to capture thoughts when coding and to keep an audit trail of decisions during theme development (37). A key part of framework analysis is development and refinement of the analytical framework which comprises codes and categories.

Codes and categories of an analytical framework can be developed 1) through open coding of data (inductively); 2) using existing theory, where codes and categories are predefined based on previous literature and or existing theories (deductively), or 3) a

combination of both approaches (38). A combination approach to develop the analytical framework was adopted where a theoretical model (Levanthal's self-regulation model (19)) was used to develop a priori codes and categories, and open coding across all transcripts was used to identify codes and categories inductively (table 2).

It was important that all interesting aspects of data were explored. A purely theoretical approach may have limited this. Open coding all transcripts was used to ensure that no data were overlooked when developing code and categories of the analytical framework (39). The self-regulation model was identified as appropriate to use to develop the a priori codes (40) as illness representations play a significant role in experiences of living with HD and could be relevant for physical activity in HD (17,18,21,22,41). To develop the a priori codes from the self-regulation model, the components that contribute to the self-regulation process were added to the framework as the a priori categories. These were namely, 'identity', 'timeline', 'cause', 'control', 'consequences', 'coping', 'evaluation' and 'integration of perceptions with memory or experiences' (table 2).

Developing codes and categories through open coding of the transcripts required logical and intuitive thinking where judgements were made about meaning, importance and relevance of issues and connections between ideas (36). Two researchers (KH, TG) double coded one transcript. The same sections of text were coded, and the same ideas about the data were expressed, although different names were given to codes. Negotiation led to renaming a small number of codes and 100% agreement was achieved. Prior to finalising the analytical coding framework, two individuals outside the research group (visiting fellows who work with people with HD) tested its usability by applying it to one transcript each.

Insert <Table 2. Analytical coding framework>

All data was indexed using the analytical framework, then charted into tables to enable the researcher to review and scrutinise data to develop key themes. Themes and subthemes were developed through a process of reviewing the data coded to the categories of the analytical framework (sample illustrated in Figure 1) where patterns and commonalities were identified across the charted data and data were checked to ensure the text fitted into the codes they had been indexed to. Overlap and repetition across codes and categories was looked for, and whether some could be collapsed together where data justified formation of an overarching theme (figure 1). Consideration was given as to whether there were better descriptions that encompassed the data in terms of developing the key themes (42).

Insert <Figure 1. Exemplar sample of the 'journey' from data extracts coded to the analytical framework, followed by grouping of categories within the framework to develop themes and examples of data coded to codes / categories of the analytical framework that were incorporated into key theme 1: evolving representations of HD and physical activity.>

Results

Three key themes were identified, namely 'the evolving representations of HD and physical activity', 'the varying social environment of the person with HD and the impact on physical activity' and 'achieving physical activity participation while coping with the nuances of HD'. A common thread throughout the three key themes is the concept of time. The themes directly link to Levanthal's self-regulation model discussed later (figure 2).

Insert <Figure 2. Illustration of the key themes mapped onto a self-regulation model (Levanthal et al. 1984)>

Key theme 1: The evolving representations of HD and physical activity

Constructs that may contribute to the formation of cognitive representations include; identity, coping, cause, consequences and control according to the self-regulation model (19). The term 'evolving representations of HD' relates to these constructs and changes over time evidenced from including participants at different stages of HD in the focus groups, but also individual experiences of HD progression and how it impacts physical activity. Perceptions of physical and psychological symptoms or 'consequences', and the impact that HD has on personal identity were important influences on physical activity participation. Ability, expectations and motivations to engage in physical activity were discussed across the focus groups. One example from an individual with early stage HD in focus group 6 is:

> I used to do all those things a real keen sportsman; football, cricket but this illness means that I'm not steady on my feet.

Reasons for being physically active included enjoyment, being outdoors, goal oriented towards maintaining activities associated with roles within the family, improving functional abilities to be able to partake in other activities such as going on holiday, training to participate in a half marathon for charity and feeling 'normal'. A spouse of someone with HD stated:

Focus group 4:

> …trying to maintain some sense of normality from doing those things is a big feature for [name] to go for that walk to be able to do it to be able to have done it is very

> …it's involving some level of physical activity which can constitute as exercise…the longer we can kind of keep it in place, better off we're going to be.

Other reasons for physical activity included social interaction, enjoying how they felt following physical activity, to 'fight back' against HD, and specific HD benefits. For example, an individual with early stage HD in focus group 5 said:

> The black cloud in the morning isn't motivating but you must get rid of it somehow...and on the treadmill you can […]. It's just a feeling of total negativity and that's my main motivator into physical activity. […] It's one of those things that I think is very important [physical activity]

Key theme 2: The varying social environment of the person with HD and the impact on physical activity

Social stigma, social support, physical activity interactions, the importance of appropriate social environment for physical activity were highlighted. Relative to the self-regulation model, experiences of physical activity influence the cognitive representations of HD, and guide coping responses and appraisal or evaluation of actions in relation to their physical activity. As such, 'coping', 'evaluation' and 'illness representations' within the self-regulation model are relevant to this theme. Participants reported feeling stigmatised when engaging in physical activity in public due to symptoms of HD. For those unaware of HD, unstable gait and chorea can cause people to be perceived as behaving inappropriately. A poignant example is described by a caregiver (spouse) in focus group 4:

> ...there became a time when he was too embarrassed to go to the gym because he felt people were looking at him. And you know, they were! I had to go and support him…

The findings clearly demonstrate that people with HD require more support with disease progression for numerous reasons i.e. cognitive, behavioural and physical consequences of symptoms. Consequential effects of these symptoms in terms of fears (fear of falling), lack of confidence in abilities, and accessing environments such as gyms and exercise classes, leisure clubs or walking groups emerged. A caregiver (spouse) explains how they deal with their fear of potential falls:

Focus group 7:

> …walking I hold her hand 99 percent of the time just in case she has a little stumble or a slip…

A clear concern across focus groups was the lack of specialist support for HD, and inconsistent provision for physical activity. Lack of specialist healthcare support has been previously identified in HD (43) but not in the context of HD and physical activity. Where support was received for physical activity through the health service people were not able to find a specific pathway to access appropriate specialised support and could be a barrier to physical activity. A pragmatic caregiver explained how they linked in with their spouse's social worker to access individualised support for physical activity. This example highlights the potential importance of the social worker role in facilitating support for physical activity in HD. The example also demonstrates that the caregiver made an objective appraisal of the situation and took action in a way they thought best to improve their spouse's situation in terms of continuing physical activity which links with the theory around 'appraisal' and 'action' underpinning the self-regulation model.

Key theme 3: Achieving physical activity participation while coping with the nuances of HD

Participants from prodromal to late stage HD described strategies they developed to engage in physical activity. Strategies such as using physical activity as a way of taking control, and adjusting expectations helped people with HD continue to be active in different ways (often with support) and adapt their physical activity from high level, to more functional activities over time, with progression of symptoms. This suggests that in line with the self-regulation model, as HD progresses an adaptive process whereby cognitive representation of the illness experience guides action planning or 'coping' responses and performance of the coping responses, followed by appraisal or monitoring of the success or failure of coping efforts. The following quotes portray how participants adapted their physical activity and set goals in line with their abilities:

Person with early stage HD, focus group 8:

> I found aerobic things too fast. I just couldn't cope with the speed mentally or physically so then they said well, this balance one is a lot slower but still does similar types of things so I went to that… I'm doing some of the exercises and I'm enjoying it.

Formal caregiver, focus group 2:

> Our goal was to push the shopping trolley round Tesco while she's walking. And she achieved that, even grabbing groceries and putting them in herself.

Key themes and modification of Levanthal's Self-regulation model

Key themes directly link to the self-regulation model (figure 2). Data coded to categories of the analytical framework developed from the self-regulation model contributed to theme formation. The self-regulation model facilitated understanding of physical activity experiences; however, it did not account for the importance of caregiver collaboration that is needed with disease progression. The original self-regulation model by Levanthal (19) suggests that individuals respond to awareness of an issue by problem solving and goal planning leading to an action plan. The action plan may be further refined over time following further appraisal of its success. In terms of responding to appraisal and evaluation of physical activity it perhaps seems obvious, that if something is not working, one will change what they are doing. In the context of a neurodegenerative disease it is not so straightforward. For someone with HD, changing how they do something may signify progression of the disease to them, although some evidence suggests that coping mechanisms as such as adjusting related to acceptance of HD have been positively related to well-being (17).

Indeed it appears that the cognitive illness representation of HD may play a role in adjusting expectations of what the individual with HD is capable of doing in adjusting to and coping with limitations caused by HD. Discussions during the focus groups highlighted cognitive linking of social, bodily and emotional experiences of HD to the impact on physical activity and how participants responded with coping strategies (or lack of ability to cope). Participants demonstrated reflexivity, sharing strategies that they had used to continue with physical activity with good effect or not so good effect, and how that changed what they did and/or how they did it, highlighting self-regulation.

In addition, the need for change in support and change in role dynamics between family members over time with HD progression was highlighted. For example, caregivers shared how they supported people with HD to reconsider and refine what they do in accordance with abilities. The caregiver appears to become a more prominent source of feedback or information in terms of informing changing representations over time and becomes more integral to the coping responses. Significantly then, the findings clearly demonstrate that with progression, caregiver input becomes vital to the daily life of the individual with HD and regulation of behaviour becomes more collaborative rather than entirely self-regulated. The regulation of physical activity behaviour in the context of HD becomes less independent over time. In light of these findings a modified self-regulation model is presented (figure 3), which accounts for the increasingly collaborative regulation of representations and physical activity participation with HD progression over time and can be used in conjunction with physical activity intervention development in HD.

The modified self-regulation model presented is still cyclical, as the findings suggest that the 'feedback loop' in terms of awareness of changes, regulation and adapting in relation to HD and physical activity is relevant. The element of time is important because over many cycles of 'self-regulation' the disease will be progressing. The arrow along the bottom of the feedback loop is separate from it; regardless of whatever is happening with 'self-regulation', time continues to pass, and progression of HD continues. As key theme two highlights, passage of time is directly linked to HD progression and the need for caregiver input related to physical activity, and increasingly collaborative regulation of physical activity is seen. As such, in the adapted model, "time" is depicted with the following text below it to show that this is a consequence of the passage of time: "increased input from caregiver; increasingly collaborative regulation of physical activity".

Insert <Figure 3. The adapted self-regulation model specific to physical activity in HD>

Discussion

HD is a devastating, progressive disease, with limited options for pharmaceutical symptom treatment. The World Health Organisation (WHO) discusses "restoring or creating a life of acceptable quality for people who suffer from neurological disorders" (44) and emerging evidence suggests physical activity engagement could have numerous positive disease specific benefits for HD including quality of life.

This original research highlights the context-specific factors associated with people with HD's engagement in physical activity, which need to be taken into account in the design of HD-tailored physical activity interventions. Similarities to previous findings in studies in MS, PD, HD were found regarding perceived importance of supporting engagement in physical activity (5,9,26,31,32,45–48). Quinn et al. (9) and Frich et al. (32) reported a lack of instances where participants had experienced the opportunity to develop a personal plan with healthcare professionals, considering their individual needs and abilities. The participants here reported a mixture of experiences in this regard, suggesting inconsistencies across the UK in terms of provision to access such support. People with HD and caregivers gave numerous reasons and motivations for physical activity participation. Motivations included perceived benefits of physical activity participation including better lifestyle and health, quality of life, maintaining independence and functional abilities for as long as possible, and opportunity for social interaction, similar to findings in MS and PD (31,46,49).

Another similarity seen in other long-term neurodegenerative conditions was the poignant motivation that being active enabled participants to fight the disease and take some control (28,50). Previous exploration of illness representations suggests that people with HD

report little personal control (18). Yet there are examples of individuals in the focus groups using physical activity as a way of empowering themselves to feel in control. This may speak to the findings of Arran et al. (18) where people feel out of control generally, and want to take back control, but also gives additional insight into how people try to do this using physical activity. The concept of 'challenges due to the physical environment' was discussed more by people with HD and caregivers for people at later stages and could be expected due to symptom progression. Psychological barriers pose a risk of decreased physical activity but are also modifiable (20). The fear of falling when being active was identified as one such psychological barrier by people with HD and caregivers from early stage onwards. The fear of falling has not been identified as a salient feature in HD; only one quantitative study of small sample size has investigated this (51). Approaches that suitably alter perceptions around the concept of falling and risk, and education around falls in HD may be fitting, and improve physical activity participation.

The concept of influencing HD related illness beliefs has previously been supported in order to improve patient well-being (17). The concept may also have a place in influencing illness representations to positively influence physical activity behaviour and overcome barriers such as fear of falling, or public stigma. This may be achieved by building confidence through education about falls, identifying and overcoming negative personal perceptions with practical solutions such as finding enjoyable ways to be active around the home, accessing one to one support as needed, and engaging with charities who can signpost to specialised support. These concepts have now informed the development of physical activity interventions (59, 60). The benefit of using framework analysis is that using the self-regulation model has identified the importance of collaborative regulation of physical activity in HD for continued participation over time. If only an inductive analysis was used rather than deductively and inductively driven analysis, this may not have emerged, but is key. The

findings have major implications for supporting and informing development and evaluation of much needed, well-defined physical activity interventions in HD. Significantly, these include caregiver importance in supporting the person with HD to engage and participate in physical activity, which over time, and with disease progression, becomes increasingly integral; in developing physical activity interventions for clinical trials, the social and familial context is therefore key to consider, as this may influence participant adherence. Differing experiences of people at various stages of HD mean that approaches to supporting physical activity require flexibility that tailors long term interventions to individual social and environmental contexts and involves discussing strategies as depicted by the findings. Such approaches are important to facilitate pragmatic physical activity participation even with progression of HD.

Facilitated by use of a qualitative method and theoretical analysis, these findings provide insight into HD specific phenomena and have informed the logic model underpinning the PACE-HD physical activity intervention. PACE-HD is a cohort study that includes an embedded randomised controlled trial of a physical activity intervention in people with early-mid stage HD (59). Logic models support successful development, implementation and evaluation of interventions through defining clear ‘inputs’, ‘activities’ and ‘outputs’ or ‘outcomes’ (8). Lack of detailed description of intervention inputs and activities make it difficult to explore reasons for lack of effect or implementation challenges (8).

The PACE-HD intervention for people with HD is delivered by physiotherapists who focus on promoting strategies to engage in aerobic exercise and physical activity (59). The program uses a disease-specific workbook and participant-coach interaction. A fundamental component of the intervention is the need for collaborative regulation that specifically considers social–contextual-disease specific conditions. This emphasises the importance of working in a collaborative way with participants, as well as understanding the progressive

nature of HD and the changing needs of the participant so that aspects such as access to healthcare and fitness facilities, social situation and adaptation to a participant's ongoing cognitive and behavioural status are all taken into account during intervention delivery.

Limitations

HD families are a hard to reach population. Individuals who could contribute a valuable perspective may not engage in research or want to join a group discussion about HD. Due to stigma of HD, potential participants may have been wary of revealing themselves. We must acknowledge the potential for selection bias in this study; participants were sufficiently interested in physical activity to volunteer participation in focus groups which were restricted to England and Wales. Our findings may not be representative of the global HD population but provide an important starting point when considering physical activity interventions in this population. We recruited a broad range of people with HD from prodromal to late stage HD, across regions with different socioeconomic status and our findings are supported by that reported in the literature, in relation to common barriers to physical activity in other neurological conditions (61, 62).

Conclusions

Understanding the impact of the nuances of HD on physical activity participation is critically important to better support physical activity in HD, given the emerging support for physical activity as a positive intervention. This research has furthered such understanding of physical activity in HD with regard to the impact of symptoms, coping responses and strategies, social environment, support needed, changing illness representations, and abilities.

Three key themes identified provide novel insight in terms of how the nuances of HD influence physical activity experiences and participation across the disease life cycle. A novel

approach of applying a theoretical model, namely the self-regulation model facilitated understanding of physical activity in the context of HD. As a result, the self-regulation model is indicated as having utility in helping to understand the experiences of physical activity in HD. However, a modified self-regulation model accounting for collaborative regulation of physical activity with progression of the disease is required. The findings provide a theoretical foundation to underpin development of a wide range of physical activity management and research interventions in HD.

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Tables

Figures

Table 1. Focus group demographics

Focus Group number	Location	Total number of participants	Participants			
			Number of people with HD		Number of caregivers (family members/formal caregivers)	
			Male	Female	Male	Female
1	Location one: Suburb, mid-England	7	3 (2 mid-late stage; 1 early stage)	1 (mid-late stage)	2 (1 father; 1 spouse)	5 (1 mother; 1 sister; 3 spouses)
2	Location two: City in Wales	5	1 (early stage)	1 (mid-late stage)	1 (spouse)	2 (1 sister;1 spouse)
3	Location two: City in Wales	5	0	2 (mid-late stage)	0	3 (formal caregivers)
4	Location three: City, northern England	4	1 (early stage)	1 (early stage)	1 (spouse)	1 (specialist HD advisor)
5	Location three: City, northern England	6	3 (early; mid; late stage)	1 (early stage)	0	2 (1 mother; 1 formal caregiver)
6	Location four: Rural southern England	5	2 (mid-stage)	1 (mid-late stage)	1 (spouse)	1 (spouse)
7	Location five: Rural, southern England	8	3 (mid-stage)	1 (mid-late stage)	1 (spouse)	1 (spouse)
8	Location five: Rural, southern England	8	4 (1 early stage; 1 gene positive)	0	0	4 (4 spouses)

Table 2. Analytical coding framework

Category	**Code**	**Code description**
Influencing Factors	Motivation	How motivation (or lack of) influences their physical activity behaviour / attitude towards physical activity.
	Environment	The influence of the environment on physical activity e.g. social / physical environment.
	Support	Different types of support discussed and how that influences physical activity behaviour.
Perceptions of physical activity	Reasons for perceptions of physical activity	Positive or negative – all perceptions of physical activity and possible reasons for those – how they have come to have those perceptions.
	Negative perceptions of physical activity	
	Positive perceptions of physical activity	
Reasons for exercising	Personal reasons for exercising	Why people with HD are active – their reasons/motivations.
	Incentives/ what encourages people to be active / exercise?	
Relationships	Caregiver/family perceptions	Impact of caregiver perception on the physical activity of people with HD.
	Interactions while exercising	Interactions whilst being active - with others / exercise trainer / family member or caregiver.
	Relationship between caregiver/family member and person with HD	The relationship between caregiver and person with HD; how this may impact on physical activity.
	Communication	Communication between people with HD and others – influence of this on physical activity.
	Social interactions	Social interactions in the context of physical activity
Stigma of HD	Public	Other people's lack of understanding or knowledge of the disease and consequences of this for physical activity.
	Family relationships	Stigma/breakdown/difficulties in family relationships because of HD
Coping	Strategies: physical activity	Specific strategies used to overcome limitations or barriers in relation to physical aNivity
	General strategies	General strategies of coping used
	Normality	What is normal to participants at different stages in terms of physical activity?
	Adapting	Adapting behaviour and or approach to physical activity because of the limitations caused by HD, accepting or not accepting change in relation to physical activity in HD
Evaluation	Reflection	Reflecting on specific experiences related to physical activity.

	Emotional experiences	Emotional responses to experiences – how do people react emotionally to certain experiences, what or how does this impact what they do in terms of PA?
	Objective response	How have people responded in a practical way to an experience they have reflected on – i.e. do they respond by troubleshooting, adapting what they do, how they do PA, ask or help or advice?
Cause	Lifestyle	Health behaviours - behaviours such as PA influencing health – what are people's perceptions of this, how does this influence their behaviour.
	Inheritance	Inheritance of the disease – chance of passing it onto children – is PA considered in terms of health behaviour to maintain health?
Consequences	Impact of symptoms	Impact of HD on physical, social, psychological function.
	Continuity	Perspective of how life will continue (also in terms of PA) in the context of the consequences of HD.
	Quality of life	Perspectives with regard to quality of life, how is it altered? Does PA mean anything in terms of improving or maintaining quality of life?
Control	Managing	Perceptions of treatment / management of HD.
	Healthcare	Contact with healthcare professionals.
	Health behaviour	Any perspectives of or beliefs in preventative health behaviours including PA.
	Control over PA	Meaning or perspectives associated with control when it comes to PA related behaviour.
Identity	Impressions of HD	How their impressions/ perceptions of the disease shape their attitudes/perceptions towards PA.
	Living with HD	How experiences of living with the disease shape their experiences and how they relate to physical activity
	Symptoms	How signs and symptoms of HD shape experiences of PA, meaning of the symptoms in the context of PA.
	Family	As a genetic disease how family members deal with HD family history.
Timeline	Change over time	Anticipated change over time and progression of the disease.
Integration of perceptions with memory or experiences	Perceptions affected by experiences	Integration of perceptions with memory or experiences.