

Using evidence-based co-design to develop a hybrid delivered exercise intervention that aims to increase confidence to exercise in people with haemophilia

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Abstract

Introduction: Due to advances in treatments, people with haemophilia (PWH) are living longer. They are not as active as the general population due to joint damage and lack confidence to be active due to concerns about further bleeds and pain. There is a need to facilitate healthy aging through promotion of physical activity (PA) and exercise. Changing patient beliefs and increasing physical literacy and confidence to move are thought to be key to helping PWH become more active.

Aim: This paper describes the development of an exercise and behaviour change intervention to improve confidence to exercise in PWH.

Methods: The 4-stage Medical Research Council framework for complex intervention development was used.

Results: Stakeholders included 17 PWH and 7 physiotherapists working in haemophilia. Seven online focus group meetings were held. The final intervention is a hybrid 12-week physiotherapist led progressive exercise programme. Classes are 45 min including Pilates, High intensity interval training and balance elements, together with discussion sessions focusing on PA recommendations, the types and benefits of different exercise styles and the effects of PA, together with the effects of aging for PWH. The COM-B model of behaviour change was used to develop the intervention.

Conclusion: Co-design helps to produce an intervention that understands the stakeholders needs. Through this process the intervention developed to incorporate not only increasing PA but also confidence to exercise. The use of behaviour change theory identified the behaviour techniques included in the intervention and aims to increase physical literacy in this population.

KEYWORDS

behaviour change, codesign, exercise intervention, haemophilia, theory of change

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1 | INTRODUCTION

Haemophilia is a rare inherited X-linked condition that affects blood clotting.¹ A major co-morbidity is that of haemarthrosis leading to haemophilic arthropathy.² Haemophilic arthropathy is associated with pain, loss of movement, reduced muscle strength together with a reduction in postural balance.²

Advances in treatment for haemophilia have led to improved life expectancy for those with the condition, to near that of the general population.³ With increasing life expectancy people with haemophilia (PWH) will experience many of the same co-morbidities of aging as the general population for example, falls, diabetes, cancer, osteoporosis, heart disease.⁴ Exercise and physical activity (PA) can prevent or reduce the impact of comorbidities in the general population and PWH should also benefit.^{5,6} Importantly, PA can reduce joint bleeds as well as provide general benefits to health and wellbeing in PWH and should be encouraged.^{7,8}

However, PWH are not as active as the general population and have reduced exercise capacity.^{9,10} They are also likely to fall more often and at a younger age.¹¹ Many PWH over the age of 40 grew up with limited treatment options when they bled.¹² Rest and avoidance of PA was often used to prevent and treat joint bleeds. As a result, many PWH have grown up hearing the message that exercise and PA may be dangerous or lack the confidence to exercise and be active.¹³

PWH face additional disease specific barriers to PA.¹⁴ Joint arthropathy leads to reduced mobility, ongoing pain and PWH worry about causing a joint bleed. This leads onto use coping strategies such as avoidance of certain situations or movements to reduce the stress on their joints and potential bleeds.¹⁵⁻¹⁷

Being advised, while growing up, not to be active and to rest to prevent bleeding has led to reduced physical literacy in the older population. There is limited evidence to suggest ways to increase confidence to exercise in PWH. Changing patient beliefs, increasing physical literacy and confidence to move are thought to be key.¹⁸ Physical literacy encompasses the motivation, confidence, physical competence, knowledge and understanding to value and take responsibility for engagement in physical activities for life.¹⁹

The aim of this paper is to describe the process underpinning the development of an exercise intervention that focuses on a behavioural change to improve confidence to exercise in PWH.

2 | METHODS

This study was registered with Oxford University Hospital National Health Service (NHS) Foundation Trusts integrated governance system (Ulysses ref 8789) as the study did not meet the requirements for formal ethics approvals. We used the updated Medical Research Council (MRC) framework for developing complex interventions.²⁰ The framework emphasises theory development, stakeholder engagement and understanding context. Involvement of key stakeholders is an essential element. Co-design is an approach that brings together

service users that is, patients and health care providers, to design and develop an intervention.²¹ This approach allows the final intervention to be produced with understanding of patient and clinician needs and has been advocated to provide more effective and sustainable interventions.²²

The development stage was divided into four steps (Figure 1).

2.1 | Step 1: (a) Identification of possible intervention components

Published literature including systematic literature reviews and clinical guidelines of exercise interventions in PWH to identify intervention components and strategies used for increasing exercise and PA were reviewed. The following databases were used PubMed/Medline, Embase, PEDro and CINAHL. Articles with the following keywords were extracted population Haemophili* OR hemophili*, interventions—PA, exercise, resistance, balance, strength*, High intensity interval training (HIIT), Mode of delivery virtual, 'face to face', 'in person, hybrid. We also used the use of Boolean operators (AND,OR,NOT). In addition, a manual search of the references of included articles was conducted. Authors also considered the National Institute for Health and Care Excellence (NICE) guidelines such as guidance on behaviour change interventions.²³

Intervention components and strategies for consideration included: falls intervention programmes, PA and strength training programmes together with modes of intervention delivery in PWH and the wider population.

The first author (S.T.) also attended virtual exercises classes run by Haemophilia Chartered Physiotherapy Association (HCPA) members to review their content.

2.2 | Step 1: (b) Identification of and engagement with stakeholders

PWH and physiotherapists working in haemophilia comprehensive care centres were identified as the two main stakeholders in the development of this exercise intervention. INVOLVE guidelines were followed and payment offered for patient time in the patient and public involvement (PPI) process.

Stakeholder meetings were facilitated by S.T., a physiotherapist working in haemophilia who had completed training with the NIHR on facilitating stakeholder groups.

PWH were invited to join the PPI group via two routes.

- The Haemophilia Society a UK-wide charity organisation for people with bleeding disorders were asked to send out a request for PWH to be involved in the development of this exercise intervention.
- PWH from other haemophilia centres were invited to participate via their physiotherapist. Physiotherapist were invited to ask their patients from an email request sent to the HCPA membership.

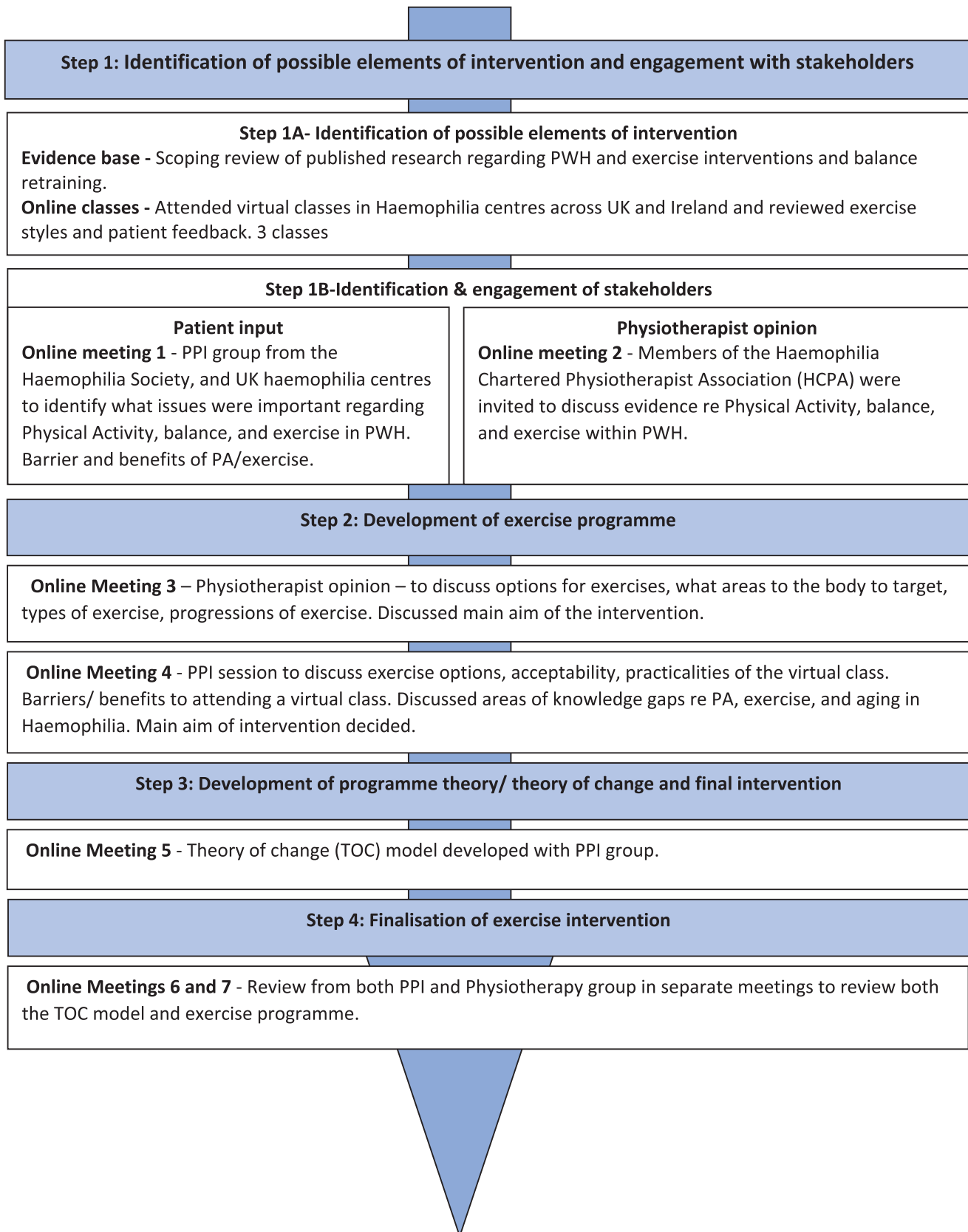


FIGURE 1 Overview of intervention development process. PPI, public patient involvement; PWH, persons with haemophilia.

The group was purposively selected to achieve a mix of haemophilia severity categories, ethnic background and age. Individuals were invited to attend a virtual session in MS Teams. An Introduction letter and briefing document were sent for each session to allow participants time to form their own opinions prior to the session. During the session participants were asked about their experiences with PA and exercise during their lifetime.

The second group of stakeholders comprised of a group of physiotherapists working in haemophilia services and was identified through the HCPA. An email was sent to all HCPA members seeking expressions of interest to participate. Members were selected to give a variety of experience within the group. An online meeting was conducted virtually on Micro Soft teams and an introductory letter together with briefing documents were provided beforehand.

Initially the research literature regarding exercise/PA in PWH was reviewed, together with physiotherapist current practise when aiming to increase PA in PWH; what exercises were used, together with methods of delivery. Members were selected to give a variety of experience within the group.

Padlet, an interactive online platform was used to help facilitate all sessions.²⁴

2.3 | Step 2: Development of exercise programme

This step comprised of separate meetings with the PPI and Physiotherapy Stakeholder groups.

The PPI meeting involved discussion regarding exercise options, which exercises would be acceptable, the practicalities of running the class and ways to reduce the barriers to being physically active and exercise. Knowledge gaps were discussed to help inform the discussion sessions within the exercise intervention.

The physiotherapists discussed options for exercise, which body areas to target, the types of exercises (e.g., strengthening, balance, aerobic) and, how these exercises could be progressed and monitored.

At the end of this stage the draft exercise content had been agreed.

2.4 | Step 3: Development of programme theory and theory of change

This step aimed to develop the programme theory and understand how the different elements of the intervention may lead to the expected outcome.²⁵

Programme theory is the formation of a model of how the intervention is meant to function. Its purpose is to encapsulate the key elements that are included within the intervention together with programme activities, inputs and outputs.

We followed the steps outlined in Figure 2 utilising information gained from published evidence and previous meetings.

PPI representatives were asked how and why the intervention may work. They were asked to consider the long-term change or outcome

being sought and to then work backwards to describe the processes and sequence of change that could lead to this outcome.

The main aim of this intervention is behaviour change, and the COM-B model was selected to underpin the intervention.²⁶ A behavioural analysis was completed in which the needs identified by the PPI group were mapped onto the COM-B domains of capability, opportunity and motivation. The theoretical domains framework was used to expand the COM-B components.

Behaviour change techniques (BCTs) from the theoretical domains framework (TDF) were identified and the list of BCTs was then reviewed using the APEASE criteria (Acceptability, Practicability, Effectiveness, Affordability, Spill-over effects and Equity) as described by Michie.²⁶

A theory of change (TOC) diagram was developed, and the PPI group reviewed it at a virtual meeting to amend and finalise the process (Figure 3).

2.5 | Step 4: Finalisation of exercise intervention

The final exercises chosen together with progressions were reviewed by a smaller subgroup of physiotherapists and the PPI group. Further amendments were made as needed. The final exercise intervention was agreed by consensus.

3 | RESULTS

3.1 | Step 1: (a) Identification of possible intervention components

We looked at the published literature to identify exercise components with either a theoretical rationale or evidence to support their provision which included:

3.1.1 | Specific types of exercises

We looked to identify specific types of exercises that were effective for improving outcomes in PWH. The latest Cochrane review investigating the effect of exercise in PWH, concluded that most exercise interventions produced an improvement in either pain, range of movement, strength and walking tolerance. These exercise interventions included strengthening with weights, stretching, exercising in water, walking and using an exercise bike. These findings are advised to be taken with caution due to the heterogeneity of outcome measures and the small number of studies. No specific type of exercise was recommended but a combination of strength training, balance, aerobic and flexibility.²⁷

Muscle mass loss is associated with haemophilic arthropathy due to recurrent joint bleeds reduced range of movement and activity.²⁸ Loss of strength has been associated with increased frailty and is a concern in an aging haemophilic population. Targeting PA and strength training has been shown to minimise these losses.²⁹ Strength and pain

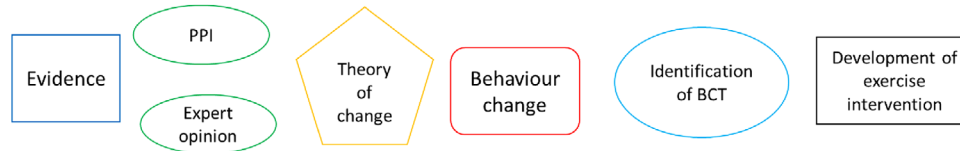


FIGURE 2 Mapping of COM-B to the theory of change model, behaviour change wheel and BCT for development of intervention. BCT, behaviour change techniques.

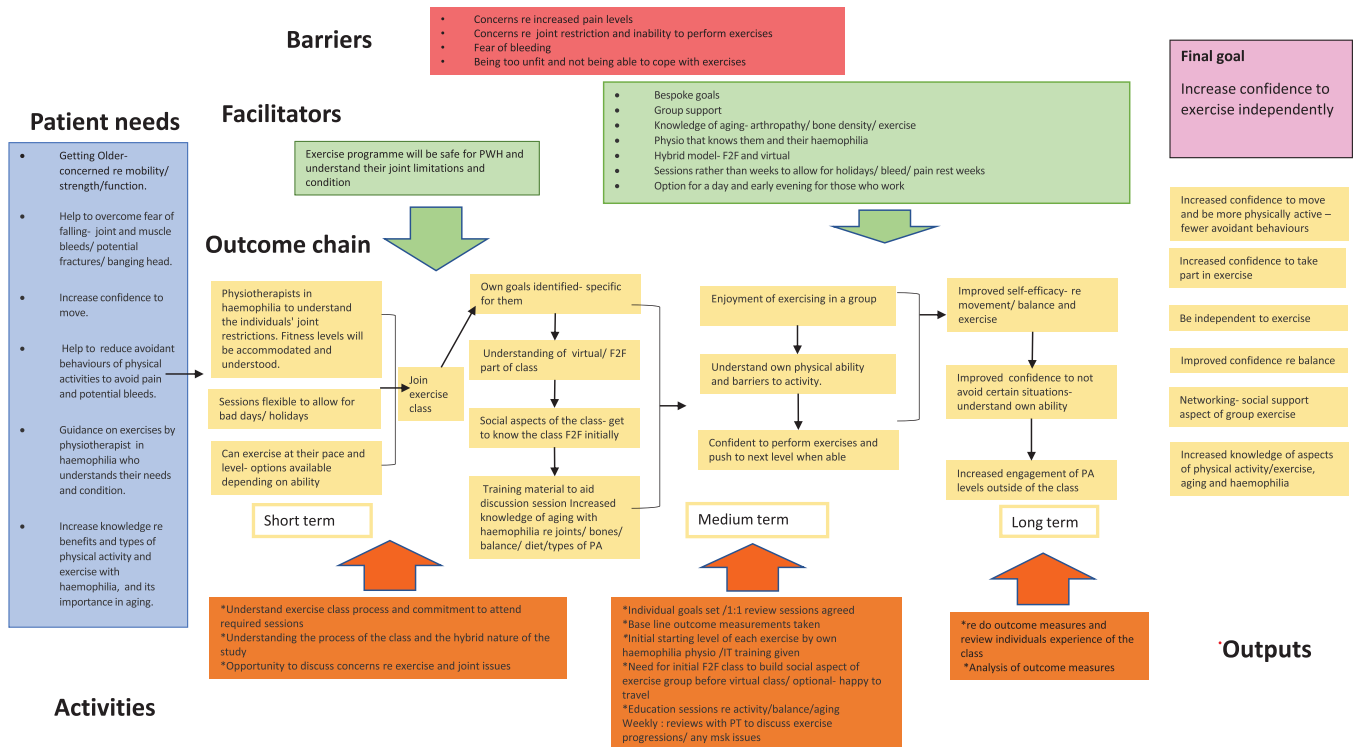


FIGURE 3 Theory of change model based on Mclaughlin et al.⁴²

have been shown to improve after an 8-week programme of resistance training.³⁰ However, the evidence from studies is of limited quality and the optimal type of exercise, dosage and timing of strengthening exercises in PWH has yet to be determined³¹

There is very little research to guide the specific exercises to be used for balance retraining in this population, rather a combination of balance, strength and endurance.³² Evidence-based strength and balance exercise programmes effectively reduce falls, increase confidence and can increase PA levels in older people.³³

3.1.2 | Mode of delivery

Exercise interventions can be delivered individually or in a group. Group classes within specific health conditions have been shown to provide not only physical benefits but also psychological benefits of peer support together with educational opportunities.³⁴ Groups could be delivered in person or virtually. Classes are already being deliv-

ered virtually for this population and with PWH often living some distance from specialist centres, this mode of delivery was important to consider.

Virtual exercise interventions have been reported as feasible in the general population.³⁵ Guo and Fussell 2022³⁵ described the challenges of delivery and interactions together with the convenience of virtual classes. Recent studies have shown that a virtual and hybrid class delivery is acceptable in PWH,³⁶ allowing for increased accessibility, especially for patients living a distance from their care centres as for many PWH.³⁷

3.1.3 | Psychosocial factors

Within haemophilia, physical literacy is a concept that is, emerging as being important.³⁸ Many PWH grew up being told not to exercise to prevent bleeds and have a poor understanding of how to be physically active, reduced confidence to be active and on what exercises could

TABLE 1 Demographic data from PPI group.

Participants	Summary of all PPI representatives involved.	Attended meeting 1	Attended meeting 4	Attended meeting 5	Attended meeting 6	Attended online exercise classes
Gender	16 males 1 female	6 males	5 males	5 males	4 males	11 males 1 female
Ethnicity	White British 15 Turkish 1 Indian 1	6 white British	4 white British 1 Turkish	4 white British 1 Turkish	3 white British 1 Indian	12 white British
Age, mean years (min/max)	55 (37–72)	50 (42–72)	52 (42–65)	52 (42–65)	55 (44–65)	54 (37–65)
Haemophilia severity type	11 Severe, 1 Moderate 5 Mild	4 severe 1 moderate 1 mild	4 severe 1 mild	4 severe 1 mild	3 severe 1 mild	7 severe 1 moderate 5 mild

TABLE 2 Demographic data physiotherapist group.

Physio group	n = 7 meeting 2	N = 3 meeting 7
Gender	3 male 4 female	3 females
Ethnicity	White British 7	White British 3
Years qualified (mean, min/max)	23 (10,35)	20 (10–35)
Years working in haemophilia (mean, min/max)	8.6 (1/17)	7.6 (1/16)
Centre type	CCC = 6 HC = 1	CCC = 3

Abbreviations: CCC, Comprehensive Centre; HC, Haemophilia Centre.

be useful. This has affected their participation in PA in adulthood.³⁹ Motivation, self-efficacy and understanding the benefits of being more physically active are all factors that lead to improved engagement across a lifetime and were identified by the PPI groups.⁴⁰

We also observed virtual exercise classes to find out what was already being provided for PWH. These classes included a class targeting those with osteoporosis, a Pilates style and class including HIIT content.

3.2 | Step 1: (b) Identification of and engagement with stakeholders

We assembled two stakeholder groups. Table 1 summarises the demographic background of the PPI group and their attendance at the meetings.

Seven physiotherapists were involved in the development of this intervention (Table 2), 7 attended the initial meeting and 3 attended the final meeting.

During meeting 1, The PPI group discussed ‘Patient needs’ which are presented (Figure 3). They were worried about aging and their primary concern was they decreased confidence to move and avoided certain activities due to pain and concerns over causing a bleed. This was identified as the main aim of the exercise programme. They were uncertain which activities/exercises were safe to do and which would



improve strength, balance and help to maintain physical function as they aged. Some members expressed a fear of falling when physically active, but this was not a primary aim as was initially thought by the research team.

3.3 | Step 2: Development of exercise programme

Meeting 3, with the physiotherapists discussed types of exercise, body areas and progressions. The group felt that a combination of strengthening, balance and aerobic exercise was important to include, especially as the main aim of the study was to increase patients’ confidence to exercise. They felt that PWH should work at a level of ‘somewhat hard’ on the rating of perceived exertion scale. Exercises should target core strength and include knee and hip extension as well as some general upper limb function to target commonly involved joints. Exercises to challenge balance, adding in dual activity exercises,⁴¹ should also be included.

The PPI group (meeting 4) discussed what exercises they felt would be acceptable to a mixed ability group, what the barriers and benefits to joining the exercise class would look like and how barriers could be mitigated. They wanted a physiotherapist who was an expert in haemophilia to take the exercise programme, they expressed negative experiences of health care professionals not experts in haemophilia causing bleeds and consequently a further reduction in confidence to be active. They wanted exercises that were targeted for their joints and abilities and to be able to adjust the difficulty to allow progression or to make it easier if their joints were painful. Patients were happy with a virtual delivery but asked for the first class to be face to face to aid communication between participants.

The exercises selected by the patient and physiotherapy groups were divided into warm up exercises, HIIT style exercises, Core strengthening and balance exercises (Figure 4). These exercise approaches were chosen to allow the participants to experience variety of exercises styles as the aim of the intervention is to increase confidence. There are options of a starting level for each exercise which can be progressed or regressed to allow patients to control their activity levels depending on joint status and pain. The class

WARM UP- 20 seconds for each exercise					
1	2		3		
	++ High knees quickly + High knee lifts		++ Single leg heel raises no support + Single heel raises holding on for balance		
Shoulder roll and full arm circles around body	Marching on the spot		Standing heel raises both legs		
					
- Shoulder rolls arms by side	- Holding on to chair smaller knee lifts.		- Heel raises both legs holding on		

HIIT Style exercises start 15 seconds increase to 40 seconds x 2 sets					
1	2		3		
++ Squats quicker and deeper squat	Recovery side steps 10 seconds		+ Press-ups full		Recovery hamstring curls 10 seconds
+ Squats increase speed			+ Press-ups on all four's hands further away		
Squats			Press ups on all fours		
- Squats smaller movement			- Press-ups against wall feet further away		
-- Squats holding on to chair			- Press-ups against wall feet nearer wall		+ add weights and speed
					+ add weights 0.5kg/1kg/2kg
					Push both arms above head
					- One arm push above head
					Marching with arm swings 10 seconds

Pilates/Core x 10 reps x 2 sets					
1	2		3	4	
	+++ Top shoulder bridge Lower leg extension leg lift- level 3			+++ 4- point kneeling opposite arms and legs lifts	
++ Hip Twist level 3 double leg lift	++ Top shoulder bridge lower leg extension level 2		++ Level 3 stretch and tap with top leg from level 2 position	++ Prone lying lift opposite arms and legs	
+ Hip Twist level 2 single leg lift knee bent	+ Top shoulder bridge Heel lift one leg		+ Level 2 heels lifted, top knee lifts	+ Prone lying lift alternate arms	
Hip Twist supine level 1	Shoulder bridge supine level 1		side lying level 1	Swimming Prone alt leg lifts	

BALANCE x 10 reps					
1	2	3	4	5	6
++ Tandem stance- eyes open / eyes closed repeat opp foot forward		++ Lift and swing the leg forward and back	++ Make circles larger	++ Knee lift larger movement, then curtsy foot off floor	++ Add dual control substitute numbers with months of the year / minutes on a clock. Then mixing the instructions.
+ Semi tandem stance eyes open /eyes closed Repeat with opposite foot forward	+ Adding neck turn left and right as you walk	+ Tap forward then swing leg behind	+ Make circles with the foot lifted from the floor	+ Knee lift forward then curtsy	+ Increase speed of movement/ larger movement
Standing feet side by side Eyes open or eyes closed for upto 15 seconds	Walking forward and backwards along a straight line on the floor	Stand on one leg and tap the opposite leg forward and back	Standing – make small circles with one leg with the foot to touching the floor	Curtsy small foot on floor	Clockface- An imaginary clockface around the patient. Patients are asked to tap a foot towards a number and return to the centre.
	- Holding on as you walk	- Tap forward and back holding on		- Curtsy holding on for extra stability.	
Discussion sessions <ul style="list-style-type: none"> • Types of physical activity and PA recommendations • Types of exercise and how can benefit PWH. • Aging and haemophilia 					

FIGURE 4 Proposed exercise programme. Green section denotes the starting level. Yellow section denotes progression options increasing in difficulty. Blue section denotes the regressions options reducing in difficulty. Pilates exercises based on APPI exercises. APPI, Australian Physiotherapy Pilates Institute.

was designed to last 45 min followed by 15 min discussion and education.

Each participant will attend 12 virtual sessions completed over a 16-week period to allow for holiday, joint bleeds or illness. Catch up 1:1 session is offered between classes if required, to progress exercises or discuss joint issues.

An overview of the exercise intervention as per TIDieR Criteria is available in Appendix 1.

3.4 | Step 3: Final theory of change model for the exercise intervention

The final TOC model developed with the PPI group is presented in Figure 3.⁴²

The main aim of the intervention was to increase confidence to exercise, but another important aim was to manage the worries regarding the function of the individual's joints. Facilitators to enable participation in the exercise intervention were: (1) Bespoke goals to account for their individual joint problems, and (2) The programme was delivered by a physiotherapist who specialised in haemophilia. PPI representatives reported poor experiences when dealing with generic health service professionals.

It is important that patients have autonomy regarding the exercise level selected each week after the 1:1 support session with their physiotherapist. Having group support was identified as important, as many had not experienced seeing other people with similar joint function and felt that this would help motivate and improve physical literacy.

4 | DISCUSSION

This paper has described the development of a complex intervention using the MRC framework. The process was iterative involving stakeholders throughout. The use of co-design helped to produce an intervention that understands patient needs and is more likely to be acceptable to both providers and users of the service, bringing together the lived experience of stakeholders and care providers with the published evidence.

There is limited evidence within haemophilia of co-design and stakeholder involvement. A recent review of co-design in healthcare revealed that this method was widely used although there is little published evidence describing the process.⁴³ Some exercise intervention studies within haemophilia have failed to produce meaningful results and one reason for this could be lack of stakeholder engagement leading to poor recruitment and large drop out.⁴⁴ A recent study which actively involved stakeholders to guide the intervention development yielded promising results.³⁶

We chose to meet with patients and clinicians separately so that patients would confidently share their thoughts and experiences. A recent study by Racine⁴⁵ evaluating stakeholder involvement observed differences in patient group user opinions depending on whether the

patient users were involved simultaneously or separately. They go on to suggest having separate groups initially to allow for independent discussion before combining user groups to allow consolidation. This process may be useful to include in future codesign interventions, as would help reduce the number of meetings during the development phase.

We found having separate PPI and professional stakeholder meetings was effective in clearly hearing each groups' perspectives. For example, initially the focus for this exercise programme development was to improve balance due to higher falls rate in PWH. However, during the initial PPI meeting it became evident that this was not a major concern. They were more concerned with the confidence to exercise and how to be physically active. They wanted support from professionals specialised in their care. The aim of the intervention was changed in response to this.

The stakeholders were concerned that their individual needs were understood and met. The variety of joint damage together with the lived experience of joint bleeds varies and having an exercise intervention with differing difficulty levels, conducted by a specialist physiotherapist was paramount for the programme to be accepted by participants. The virtual component should help with inclusion as many patients live a long way from treatment centres as would limiting classes to 45 min.

Physiotherapy interventions targeting health promotion for example, increasing PA, require that behaviour change is included.⁴⁶ The NICE recommends that interventions are underpinned by theoretic models. In this intervention we integrated the theory of behaviour change. We mapped the TDF to the COM-B model and behaviour change wheel (BCW). This allowed the intervention to include activities aimed at reducing barriers to participation, improving retention in the exercise programme and to promote increased confidence to be more physically active and exercise. We also included a theoretical basis for the psychological aspect of the programme by drawing on the concept of physical literacy. This has been identified as an effective theoretic model for enhancing PA interventions for sustaining PA across a lifetime.

5 | STRENGTHS AND WEAKNESSES

The strength of this work is that we used a well described theory-based programme development that has been shown to lead to a better understanding of patient needs, the intervention programme development, and outcomes. We followed a logical approach using the MRC framework and use of PPI from the beginning of the project. Adopting a recognised theory of change model ensures justification of the chosen intervention components and transparency throughout. The patient participants severity types were representative of haemophilia severity prevalence making the co-production output potentially relevant to many PWH.

However, it was challenging to engage voices from patients from all ethnicities and backgrounds and despite our efforts to foster an inclusive approach to actively seek out equality and diversity in participants

we only managed to recruit a small number of service users with limited ethnic diversity so views may not reflect all PWH.

6 | CONCLUSION

This paper describes the process of co-designing an exercise intervention for PWH to increase confidence to exercise, using the MRC framework for complex intervention design. The use of codesign and the development of a theory of change helped to identify the participants needs and factors to help facilitate change. The use of the outcome chain identifies how the behavioural change may occur in this exercise intervention, using the COM-B and BCTs. This framework identifies the needs of both patients and health care providers to produce a potentially effective and implementable intervention, but further evaluation is needed.

AUTHOR CONTRIBUTIONS

Stephanie Taylor designed the study and conducted the development and final design of the intervention and drafted the first draft. Esther Williamson and David Stephensen provided expert oversight for the study design and development and reviewed all versions of the manuscript. Karen Barker provided expert review of the final development process and manuscripts.

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CONFLICT OF INTEREST STATEMENT

All authors stated that they had no interested which might be perceived as posing a conflict or bias.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ETHICS STATEMENT

Research ethics committee (REC) approval was not required for the preparation of this manuscript. This study was registered with the Oxford University Hospital NHS Foundation Trusts integrated governance system.

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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