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ORIGINAL ARTICLE

Fitness enhances psychosocial well-being and self-confidence in young men with hemophilia: Results from Project GYM

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Abstract

Introduction: Contemporary hemophilia care supports physical activity, its benefits being well recognized. Despite recognition of the psychological challenges encountered by people with hemophilia, little is known about the psychological impact of physical fitness in this population.

Aim: To identify changes in psychological well-being in young men with hemophilia through participation in a gym program.

Methods: This observational feasibility study of a 6-month gym participation program used validated questionnaires pre- and poststudy to evaluate motivation to exercise, physical activity levels, self-efficacy, self-esteem, and quality of life. Individual audio-recorded interviews about study participation and impact were transcribed verbatim and analyzed for recurring themes using thematic analysis.

Results: Nineteen participants aged 18–25 years with hemophilia A or B (all severities ± inhibitor) consented to the study; two were lost to follow-up. There was a shift in motivation to exercise as shown by the Stages of Change grouping moving from contemplation to action and maintenance phases ($p = 0.03$). Self-efficacy overall scores showed a trend ($p < 0.06$) towards improvement. Median self-esteem scores improved from 22 (range 12–30, $n = 19$) to 25 (range 13–30, $n = 17$), a statistically significant change ($p = 0.02$). Three participants recorded scores below the accepted normal range before study, of whom two improved at study end. The key themes identified from the interviews were: fear, self-confidence, "being normal," pain, weight loss, ability, getting fitter.

Conclusions: The psychological wellbeing of young men with hemophilia improved during this study. This may have been related to participating in a gym-based, physical exercise program.

KEYWORDS
Fitbit, gym, hemophilia, psychological wellbeing, self-confidence

Essentials

- Psychological benefit of physical fitness in people with hemophilia (PWH) is poorly understood.
- This study aimed to enable PWH to define, meet and maintain individual fitness goals.
- Themes identified were fear, self-confidence, 'being normal', pain, weight loss, ability, and getting fitter.
- Psychological wellbeing (motivation self-efficacy and self-esteem) improved during the study.

1 | INTRODUCTION

Life expectancy for people with hemophilia (PWH) has increased because of advances in treatment. Alongside this, there is an emergence of age-related disorders such as cardiovascular disease, overweight, obesity, and cancer at the same rate as the general population.[1-3] The UK government's public health strategy recommends that all adults are physically active for 150 min of moderate intensity activity each week[4] and encourages participation in sport.[5]

Improved physical health has been shown to positively impact mental health.[6] A large cross-sectional study demonstrated that physical exercise was significantly associated with improved self-reported mental health with a 22.3% and 20.1% reduction in mental health issues in team sports and aerobic gym activities, respectively.[7]

Contemporary hemophilia care supports physical activity "to promote physical fitness and normal neuromuscular development, with attention paid to physical functioning, healthy body weight, and self-esteem."[8] The physical benefits of exercise in PWH are well recognized,[9] despite the psychological challenges they face.[10] However, there are few data on the psychological determinants and outcomes of physical fitness in PWH.

In the SO-FIT study, boys (aged 8–17 years) self-reported good physical functioning overall but impairment in the domains of "friends" and "sports and school."[11] In a UK study of 84 children with hemophilia (mean age 11.5 years), 95% participated in sport and reported better physical performance and quality of life (QoL) than the boys who did not.[12] Adults reported less sports participation, which was attributed to physical condition or fear of injury.[13] Despite reporting motivation to "get fit" and "lose weight," young adults also report that they are not reaching their full ability because of perceived fears about bleeding and injury.[14] This concurs with Buxbaum et al., who report lower self-efficacy scores in adolescents with hemophilia than in controls and suggests that this is related to perceived barriers in physical activity.[15] Taylor et al. reported that UK adults with hemophilia wished to be active but spent many hours in sedentary states because of pain or the fear of bleeding.[16] These studies suggest that although, at this time, children with hemophilia engage in physical activities and sports, adolescents and adults report perceived challenges in participation because of their hemophilia. The psychological benefits of exercise and fitness are rarely reported by PWH because of previous personal history, a lack of self-confidence, and the perceived ongoing risk of injury.

The objectives of Project GYM were to identify strategies that would enable PWH to define, meet and maintain their individual fitness goals. The principal efficacy and safety results of Project GYM have been reported elsewhere[17]; the psychological findings of this study are reported in this paper.

2 | METHODS

This feasibility study recruited young men, aged 18–25 years, with hemophilia A or B (all severities ± inhibitor), via three London hemophilia centers. Participants attended a Project GYM induction session and were given free access to a central London gym and a Fitbit activity tracker to wear. As a feasibility study, a pragmatic number of 20 participants was intended with 10 in each of two randomized subgroups: personal trainer versus "gym only" (i.e., self-trained). Following randomization using a sealed envelope system, half of the participants were given fully funded access to a personal trainer. The study received Health Research Authority and ethical approval (IRAS project ID: 241384; REC reference 18/WA/0179).

During the gym induction, participants completed baseline questionnaires to evaluate motivation to exercise (the Stages of Change Questionnaire),[18,19] physical activity (International Physical Activity Questionnaire [IPAQ]),[20] self-efficacy (Self Efficacy to Manage Chronic Disease Scale),[21] self-esteem (Rosenberg's Self-Esteem Scale),[22] and QoL (EQ-5D).[23] These were repeated 6 months later alongside a semistructured interview, either face to face or by telephone, to explore their reasons for joining the study, the facilitators or barriers to completing the program, and any other highlights or concerns. The interviews followed an initial topic guide that was adapted to reflect the views of earlier respondents. Interviews were recorded, transcribed verbatim, and analyzed using thematic analysis.

2.1 | Analysis

This study was not designed to compare differences between the two groups but as a feasibility study to evaluate motivation to exercise and its impact on physical activity, self-efficacy, self-esteem, and QoL. The quantitative data were analyzed using SPSS (version 25; SPSS Chicago, IL). Descriptive data are shown as median ± standard deviation, median, and range (minimum–maximum). Bootstrap confidence intervals and standard deviations were computed using R (seed = 129,791, number of resamples = 100,000). Bootstrapping

was used to estimate the summary statistics for this population from a small (yet representative) sample group.

Differences in efficacy endpoints (changes in scores start vs. end) were tested in R using Pearson's chi-squared test and Wilcoxon signed-rank test with continuity correction. A $p < 0.05$ was considered statistically significant.

The qualitative data from the interviews were analyzed using NVIVO 12 for Mac. Each interview was read independently by two authors (K.K., S.F.), and comments were identified and coded using thematic analysis.[24] All coded comments were then reviewed and developed into overarching themes and agreed by all authors.

3 | RESULTS

Nineteen participants median age of 22.1 (18.1–24.1) years from a wide ethnic mix were recruited; 14 had hemophilia A, 12 were severely affected, four had mild, and three had moderate hemophilia. The majority of participants all with severe hemophilia (13/19, 68.4%) received prophylactic factor replacement (three with extended half-life products), there were no current inhibitors; full patient demographic data are reported elsewhere.[17] Nineteen participants commenced the study and 6-month follow-up questionnaires were completed by 17 participants; 10 participants agreed to the final recorded interview; five of these were face to face and five by telephone lasting a median 10 min (range 5.5–24.5). Of the remainder, six did not respond to interview invitations, and one left the United Kingdom for a gap year and was not able to be contacted. There were no discernible differences between those who were interviewed and those not in the overall data analysis.

3.1 | Motivation to exercise

During the course of the study, there was a significant shift in motivation to exercise as shown by Stages of Change grouping (Table 1). At baseline, participants appeared to be motivated to change their exercise levels, with one in the contemplation phase, four in preparation, three in action phase, and 11 in the maintenance phase. By study end, 16 were in maintenance and one was in the action phase. Data collected using the IPAQ short form showed that activity levels increased in seven participants and decreased in two, with no change in eight; follow-up data were missing for two participants. The number of participants with a high level of physical activity (20 min vigorous activity on at least 3 days, or 7 or more days of walking for at least 30 min per day in a week [as defined by the IPAQ]) increased from eight to 13. Those in the low (no moderate or vigorous activity) and moderate (3 days of 20 min of activity or 5 days of walking for 30 min) categories decreased from four to zero and seven to four, respectively. Those participants with access to a personal trainer attended the gym more regularly; however, there was no statistically significant difference in IPAQ score between the two groups.

TABLE 1 Global changes in efficacy endpoints

Measure	Statistic	Data		Test	*p* value
		Prestudy ($n = 19$)	Poststudy ($n = 17$)		
Stages of change	Change in grouping (start vs. end)	Precontemplation (0) Contemplation (1) Preparation (4) Action (3) Maintenance (11)	Precontemplation (0) Contemplation (0) Preparation (0) Action (1) Maintenance (16)	Pearson's $\chi^2 = 6.84$, df = 2	0.03
IPAQ	Change in score (start vs. end)	Low (4) Moderate (7) High (8)	Low (2)[a] Moderate (4) High (13)	Pearson's $\chi^2 = 2.68$, df = 2	0.27
Self-efficacy	Change in score (start vs. end)	Median = 49 Range = 19–60	Median = 52.5 Range = 25–60	Wilcoxon signed-rank test with continuity correction $Z = 22.5$	0.06
EQ-5D index score	Change in score (start vs. end)	Median = 0.837 Range = 0.414–1.000	Median = 0.819 Range = 0.512–1.000	Wilcoxon signed-rank test with continuity correction $Z = 26$	0.56
EQ-5D VAS	Change in score (start vs. end)	Median = 75 Range = 15–100	Median = 80 Range = 45–98	Wilcoxon signed-rank test $Z = 24.5$	0.05
Self-esteem	Change in score (start vs. end)	Total score median = 22, range = 12–30	Median = 26 Range = 13–30	Wilcoxon signed-rank test with continuity correction $Z = 11$	0.02

[a]Assigned values because of noncompletion.

3.2 | Self-efficacy

At baseline, the median self-efficacy score (scored from 0 to 10) for participants was 8.17 (range 3.17–10.0, $n = 19$); by study end, the scores had improved to 8.75 (range 4.17–10.0, $n = 16$). Although nonsignificant, the overall self-efficacy scores increased from baseline to study end (Table 2). Two subquestions ("confidence in keeping the physical discomfort or pain of disease from interfering with the things the patient wishes to do" and "confidence in keeping the emotional distress caused by disease from interfering with the things the patient wishes to do") showed a significant increase ($p < 0.05$) in the poststudy survey results.

3.3 | Self-esteem

At baseline, the median self-esteem score using the Rosenberg's Self-Esteem Scale (range 0–30) was 22 (12–30, $n = 19$), with three participants recording scores below the reported normal range (15–25), suggesting low self-esteem. At study end, the median self-esteem score was 25 (range 13–30, $n = 17$). Twelve participants showed significant improvements ($p = 0.02$) and only one recorded a score below the normal range. Global changes in self-esteem responses are summarized in Table 3.

The baseline dataset indicated a significant ($p < 0.01$) correlation between self-esteem and self-efficacy that was maintained at study end (baseline Spearman's rank correlation coefficient (r) 0.65, $p \leq 0.01$, $n = 18$; study end 0.90, $p \leq 0.01$, $n = 14$).

3.4 | Quality of life

QoL at baseline was reported as good (median EQ-5D index value 0.84, score range 0–1 where 1 corresponds to full health) and showed no significant change over the study duration (study end median value 0.82 [$p < 0.56$]), although the EQ-5D Visual Analog Scale (VAS) score (range 0–100) where 100 represents the "best" score increased significantly between study start and end (Wilcoxon signed-rank test, $Z = 24.5$, $p = 0.05$). Overall, scores remained the same for six participants, worsened for four, and improved for seven. Clinically meaningful change[25] occurred in six participants with improved scores using the VAS. Table 4 shows the number and proportions of those reporting perfect health versus problems within the domains of the EQ-5D. One participant reported more problems with mobility at study end; however, reductions were seen in the number of participants reporting problems in both the pain/discomfort and anxiety/depression domains. For the pain domain, two with severe and two with moderate hemophilia reported "slight" pain on the EQ-5D; this did not correlate with their recent hemophilia joint health scores (median 1, range 0–7 [$n = 7$] in the moderate and 3, range 0–13 [$n = 12$] in the severe hemophilia groups). There was improvement with participants ($n = 17$) recording pain (no pain or slight pain) at follow-up (7 vs. 9 and 8 vs. 5, respectively). Two participants recorded moderate pain at both study start and end, and one reported slight pain becoming moderate, unrelated to bleeding. There was a significant correlation between EQ-5D index and self-esteem in both prestudy and poststudy data ($p = 0.02$ vs $p = 0.01$); using the VAS data, there was a significant correlation with self-esteem only in the poststudy data ($p = 0.08$ vs $p = 0.01$; Table 5).

3.5 | Interview data

Key themes identified from the interviews were: fear and self-confidence, being normal, pain, weight loss, ability, getting fitter (Table 6).

Fear often arose from parental concern about the safety of sport and exercise. Some participants reported this had limited their own willingness to participate, which in turn affected self-confidence. Being able to participate in a study that enabled "safe" gym use improved confidence:

> I feel more happier and more confident in general – it's made me really happy … it has really changed me, I'm going to be a different person because of this.
>
> (21 year old, severe hemophilia A)

Improved confidence led to comments about being perceived as "normal" and able to participate fully in activities alongside peers.

> I'm certainly more aware that I can do as much as the next person can.
>
> *(23 year old, severe hemophilia A)*

> I don't think people with hemophilia should be any different.
>
> (24 year old, severe hemophilia B)

Those with pain reported being able to differentiate pain caused by exercise rather than hemophilia. They also stated that they reduced treating what they thought might be early bleed symptoms and, subsequently, analgesia use.

> I have sore ankles and now if I have done a certain amount of steps in the day I can think …that's activity not bleeds so I'm not taking pain killers just for the sake of it.
>
> (24-year-old, severe hemophilia B)

With regard to becoming fitter and healthier, participants reported feeling physically better in themselves, which improved self-confidence. This included weight loss related to increased exercise "using the gym, walking and running more" or "drinking less alcohol as part of being healthier." Several participants commented on how using the gym had made them feel more like their peers and siblings and able to participate in more team sport:

> The last 4 months, I've started playing a lot more football and I feel much better. Among my friends, we play three times a week, so it's just like a group interest. It's just something I enjoy, so I play as much as I can.
>
> (23 year old, severe hemophilia B)

The most commonly expressed reasons for joining the study were a desire to become fitter and healthier ($n = 10$), to gain muscle mass ($n = 7$), to gain confidence in their ability to use the gym and to exercise ($n = 4$), and to lose weight ($n = 3$). These goals remained the same at study end, although several participants also expressed the desire to use the gym more often.

4 | DISCUSSION

In this study, we have demonstrated motivation to increase physical activity levels. We found no significant change overall in self-efficacy, although two questions in the Self Efficacy to Manage Chronic Disease Scale indicated significant improvements at study end in questions relating to participants' confidence in keeping the emotional distress and the physical discomfort or pain of hemophilia from interfering with the things they wish to do. Pinto et al. suggest that emotional distress in PWH is a "cause or consequence of disease or treatment" and that psychological assessment and intervention is warranted in hemophilia care.[19] Our study suggests that physical activity may be a useful strategy for reducing emotional distress and pain. Pain remains a central issue for PWH because of acute bleeding and arthropathy and is associated with emotional distress.[26] Participants in our study were well treated with intensive prophylaxis and had good joint health,[17] yet some still reported pain and considered this a reason to limit participation in physical activity. Conversely, one participant reported that physical activity improved his pain:

> When I started exercising … the pain disappeared, when I didn't come the pain started to appear back, so it's really important to come to the gym to avoid that … it really helped me not to have so much pain as I used to.
>
> (21-year-old, severe hemophilia A)

QoL scores at the start of this study were rated as good, with a median EQ-5D index score of 0.84 of a maximum permissible score of 1.0. We found no significant change in the EQ-5D index score over the course of the study, although there was significant improvement in the EQ-5D VAS score, a measure of participants' overall assessment of their health. The lack of impact on the index score is not surprising: because this group had received intensive, often primary, prophylaxis (data not shown), it may be that EQ-5D is insufficiently sensitive to show an improvement in QoL. The EQ-5D is a generic assessment instrument, and Payakachat et al. recommend using a condition-specific measure alongside it.[27] Such an approach may have demonstrated a greater effect. However, the EQ-5D index scores were similar to those reported by O'Hara et al., who recorded

TABLE 2 Global changes in self-efficacy responses ($n = 16$)

Question: How confident are you that you can	Baseline	Poststudy	*Z*	*p* value
1. Keep the fatigue caused by your disease from interfering with the things you want to do?	Median: 8; [min max]: 3 10 SD: 0.750; CI (high low): 9 7	Median: 8.5; [min max]: 4 10 SD: 0.790; CI (high low): 10 7	16	0.07
2. Keep the physical discomfort or pain of your disease from interfering with the things you want to do?	Median: 8; [min max]: 3 10 SD: 0.898; CI (high low): 9 5	Median: 8; [min max]: 4 10 SD: 0.592; CI (high low): 10 8	7	0.01
3. Keep the emotional distress caused by your disease from interfering with the things you want to do?	Median: 8; [min max]: 1 10 SD: 1.756; CI (high low): 10 5	Median: 9; [min max]: 3 10 SD: 0.763; CI (high low): 10 7	6.5	0.04
4. Keep any other symptoms or health problems you have from interfering with the things you want to do?	Median: 8.5; [min max]: 3 10 SD: 0.838; CI (high low): 9 6	Median: 9; [min max]: 4 10 SD: 0.741; CI (high low): 10 7	22.5	0.20
5. Do the different tasks and activities needed to manage your health condition so as to reduce your need to see a doctor?	Median: 9.5; [min max]: 3 10 SD: 0.719; CI (high low): 10 8	Median: 8.5; [min max]: 4 10 SD: 0.505; CI (high low): 9 8	44	0.72
6. Do things other than just taking medication to reduce how your illness affects your everyday life?	Median: 9; [min max]: 2 10 SD: 0.749; CI (high low): 10 7	Median: 9; [min max]: 4 10 SD: 0.653; CI (high low): 10 7	36.5	0.87
Total score	Median: 49; [min max]: 19 60 SD: 5.194; CI (high low): 56 39	Median: 952.5; [min max]: 25 60 SD: 3.092; CI (high low): 55 45	22.5	0.06

Note: Bootstrap confidence intervals and standard deviations computed using R (seed = 129,791, number of resamples = 100,000).

TABLE 3 Global changes in self-esteem responses

Question	Baseline	Poststudy	*Z* score	*p* value
1. I feel that I am a person of worth, at least on an equal plane with others	*n* = 18 Median (min max): 3 (2 3); SD 0.141; CI (high low) 3 2.5	*n* = 18 Median (min max): 3 (2 3); SD 0.142; CI (high low) 3 2.5	5	0.99
2. I feel that I have a number of good qualities	*n* = 17[a] Median (min max): 3 (2 3); SD 0.289; CI (high low) 3 2	*n* = 17 Median (min max): 3 (2 3); SD 0.179; CI (high low) 3 2	2	0.77
3. All in all, I am inclined to feel that I am a failure	*n* = 18 Median (min max): 2 (0 3); SD 0.246; CI (high low) 3 2	*n* = 18 Median (min max): 3 (1 30); SD 0.445; CI (high low) 3 2	0	0.09
4. I am able to do things as well as most other people	*n* = 18 Median (min max): 2 (1 3); SD 0.357; CI (high low) 3 2	*n* = 18 Median (min max): 3 (2 3); SD 0.246; CI (high low) 3 2	10	0.06
5. I feel I do not have much to be proud of	*n* = 17 Median (min max): 2 (1 3); SD 0.292; CI (high low) 3 2	*n* = 17 Median (min max): 3 (1 3); SD 0.180; CI (high low) 3 2	12	0.04
6. I take a positive attitude toward myself	*n* = 17 Median (min max): 2 (1 3); SD 0.183; CI (high low) 3 2	*n* = 17 Median (min max): 3 (1 3); SD 0.400; CI (high low) 3 2	9	0.1
7. On the whole, I am satisfied with myself	*n* = 17 Median (min max): 2 (1 3); SD 0.458; CI (high low) 3 1	*n* = 17 Median (min max): 3 (1 3); SD 0.470; CI (high low) 3 2	13.5	0.28
8. I wish I could have more respect for myself	*n* = 16 Median (min max): 2 (1 3); SD 0.437; CI (high low) 2 1	*n* = 16 Median (min max): 2 (0 3); SD 0.324; CI (high low) 3 1	3.5	0.13
9. I certainly feel useless at times	*n* = 17 Median (min max): 2 (0 3); SD 0.440; CI (high low) 3 1	*n* = 17 Median (min max): 2 (1 3); SD 0.482; CI (high low) 3 2	4	0.07
10. At times I think I am no good at all	*n* = 17 Median (min max): 2.5 (1 3); SD 0.432; CI (high low) 3 2	*n* = 17 Median (min max): 3 (1 3); SD 0.400; CI (high low) 3 2	9	0.82
Total score	*n* = 18 Median (min max): 22 (12 30); SD 1.547; CI (high low) 25.5 19.75	*n* = 15 Median (min max): 26 (13 30); SD 2.142; CI (high low) 28.5 20.5	11	0.01

Note: Bootstrap confidence intervals and standard deviations computed using R (seed = 129,791, # resamples = 100,000).

[a]Missing data where numbers are <18.

TABLE 4 Numbers and proportions reporting perfect health versus problems within the domains of the EQ-5D

	Mobility		Self-care		Usual activities		Pain/discomfort		Anxiety/depression	
Level	Baseline (*n* = 19)	Poststudy (*n* = 17)	Baseline (*n* = 19)	Poststudy (*n* = 17)	Baseline (*n* = 19)	Poststudy (*n* = 17)	Baseline (*n* = 19)	Poststudy (*n* = 17)	Baseline (*n* = 19)	Poststudy (*n* = 17)
Number reporting no problem (level 1)	13 (68%)	10 (59%)	18 (94%)	16 (94%)	14 (74%)	12 [71%)	8 (42%)	9 (53%)	9 (47%)	11 (65%)
Number reporting some problem (levels 2–5)	6 (32%)	7 (41%)	1 (6%)	1 (6%)	5 (26%)	5 (29%)	11 (58%)	8 (47%)	10 (53%)	6 (35%)
Change in numbers reporting problems	+1		0		0		−3		−4	

TABLE 5 Regression analyses on EQ-5D and self-esteem score

	EQ-5D Index		Self-esteem				
	Median	Range	Median	Range	Spearman's rank correlation coefficient	*p* value	*n*
Baseline	0.837	0.414–1.000	22	12–30	0.545	0.02	18
Poststudy	0.786	0.512–1.000	26	13–30	0.646	0.01	15
	EQ-5D VAS		**Self-esteem**				
	Median	Range	Median	Range	Spearman's rank correlation coefficient	*p* value	*n*
Baseline	75	15–100	22	12–30	0.425	0.08	18
Poststudy	80	45–98	26	13–30	0.633	0.01	15

Note: Missing/incomplete responses: prestudy (1), poststudy (4).

TABLE 6 Themes and supporting quotes

Theme	Example quotes
Fear and self-confidence	"I was quite afraid, I really wanted to do a sport or gym to be fitter and healthier" (21 year old, severe hemophilia A)
	"This will sound a bit weird … but you are not the same as everyone else – don't try to do what others do, be conscious that you don't have to do this" (23 year old, severe hemophilia A)
	"My mum and dad refused to let me do sport [because of hemophilia], I like it and I want to be good at it" (22 year old, severe hemophilia A)
	"I feel more happier and more confident in general – it's made me really happy … it has really changed me, I'm going to be a different person because of this" (21 year old, severe hemophilia A)
	"I feel like it has gone up, the confidence and stuff" (20-year-old, moderate hemophilia B)
Being normal	"I'm certainly more aware that I can do as much as the next person can" (23 year old, severe hemophilia A)
	"I don't think people with hemophilia should be any different" (24 year old, severe hemophilia B)
	"I can train hard and I can be just as good as you, if not more" (22 year old, severe hemophilia A)
Pain	'I have sore ankles and now if I have done a certain amount of steps in the day I can think … that's activity not bleeds so I'm not taking pain killers just for the sake of it" (24 year old, severe hemophilia B)
Weight loss	"I have lost weight by going to the gym" (23 year old, severe hemophilia A)
	"It's not weight – I eat health [sic] now, I haven't been drinking (alcohol)… it's all just coming together" (22 year old, severe hemophilia A)
Ability	(One participant took a physical therapy course – he is now a qualified physical therapist): "I have found my niche in climbing, I want to do that and more outdoor activities … this is where my career is" (22 year old, severe hemophilia A)
	"I had some problems with my ankle – we made some changes to the exercises so I could carry on" (21 year old, severe hemophilia A)
Getting fitter	"The last 4 months I've started playing a lot more football and I feel much better" (23 year old, severe hemophilia B)
	"I feel a bit better to be honest, I can do more, I can walk …well I can run longer and do more, I don't feel that easily out of breath" (20 year old, moderate hemophilia B)

a mean score of 0.87 for those with no target joints in a European study.[28] Carroll et al. showed that QoL is only affected in those with two or more target joints and those with a high frequency of joint pain or surgery,[29] which was not seen in our study cohort.[17]

Physical ability and body image are known to affect self-esteem and are reported as lower in young men with hemophilia than their peers.[15] Limperg et al. showed that young adult Dutch men with bleeding disorders had significantly lower self-esteem than their peers and postulated that this was related to reduced physical ability and professional success.[30] In a Turkish study of adolescents with hemophilia, treated on demand, with poor body image from bleeds including joint and muscle damage, the importance of psychological

support to address image was recognized.[31] Our study demonstrated a significant improvement in participants' self-esteem scores. There was also an improvement in response to the sub question "I feel I do not have much to be proud of," suggesting that being able to participate in "normal" gym activities (as reported by a 22-year-old participant: "I can train hard and I can be just as good as you, if not more") improves self-esteem through fitness. Exercise programs are known to "foster a sense of social belonging, motivation and efficacy" and may enhance successful adherence to exercise participation,[32] which in turn affects body image.[33]

The participants wore a Fitbit activity tracker device and were able to monitor their own step counts. In a study of PWH, Carrasco et al. reported that Fitbit use alone did not increase motivation for physical activity and that additional motivational strategies were necessary.[34] Although uploading of activity data for study analysis was poor across the cohort, individuals reported benefit from step counts in encouraging them to move more:

> It's had an impact …how many steps I'm doing and thinking 'oh wow, I really don't' move much in the day' (office worker), so I force myself to go and get my activity levels to peak.
>
> (24 year old, severe hemophilia B)

Participants also reported additional benefits of less fear, reduced pain, weight loss, and improved confidence, perhaps from seeing success in their own gym activity. PWH who practice any type of physical activity tend to report decreased emotional distress and depression compared with those who are sedentary.[35] Working with a personal trainer did not appear to impact psychological outcomes, although those who did attended the gym more frequently, potentially suggesting better motivation, which may affect long-term outcome.

We recognize that our study is not fully generalizable. The sample size is small and so the statistical assessment is underpowered, although there are some significant findings of tests within tests. The participants were selected from three large teaching hospitals where intensive primary prophylaxis had been the standard of treatment for most; thus, joint health and QoL were good at study start.[17] No clinical recommendations can be made for the global hemophilia community. The age range of study participants (median 22.1 years) may have affected motivation and adherence to use the gym and to participate fully in the study, which led to low levels of poststudy interviews and follow-up. Nevertheless, this study has demonstrated the feasibility of a tailored physical training program and suggests that encouraging young men with hemophilia to engage in a physical activity may be associated with improvements in psychological well-being. The improvement we have seen in self-efficacy may be related to the known effect of exercise on reducing pain, helping weight loss, and improving body image.

Gym participation is considered a "normal" and wished-for activity for young men with hemophilia.[14] Further study with groups of older participants, those with other bleeding disorders as well as women in gyms are required, as is a need to assess and treat psychological health as part of routine hemophilia care using generic and hemophilia-specific tools.

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AUTHOR CONTRIBUTIONS

Kate Khair, Mike Holland, and Paul McLaughlin designed the study, collected and analyzed the data, and wrote the first draft of the paper. Simon Fletcher analyzed the qualitative data. All authors critically reviewed revisions of the manuscript and agreed its final version.

RELATIONSHIP DISCLOSURE

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