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ORIGINAL ARTICLE

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Similar sports participation as the general population in Dutch persons with haemophilia; results from a nationwide study

Olav Versloot[1] | Erna C. van Balen[2] | Shermarke Hassan[2] | Saskia E.M. Schols[3,4] | Frank W.G. Leebeek[5] | Jeroen Eikenboom[6] | Michiel Coppens[7] | Lize F.D. van Vulpen[1] | Cees Smit[2] | Mariëtte H.E. Driessens[8] | Janjaap van der Net[9] | Samantha C. Gouw[2,10] | Kathelijn Fischer[1] | on behalf of the Haemophilia in the Netherlands 6 steering committee[1]

[1] Van Creveldkliniek, University Medical Center Utrecht, Utrecht, The Netherlands

[2] Department of Clinical Epidemiology, Leiden University Medical Center, Leiden, The Netherlands

[3] Department of Hematology, Radboud University Medical Center, Nijmegen, The Netherlands

[4] Hemophilia Treatment Center Nijmegen-Eindhoven-Maastricht, Nijmegen, The Netherlands

[5] Department of Hematology, Erasmus MC, University Medical Center Rotterdam, Rotterdam, The Netherlands

[6] Department of Internal Medicine, Division of Thrombosis and Hemostasis, Leiden University Medical Center, Leiden, The Netherlands

[7] Department of Vascular Medicine, Amsterdam University Medical CentersUniversity of Amsterdam, Amsterdam, The Netherlands

[8] Netherlands Haemophilia Patient Society (NVHP), Nijkerk, The Netherlands

[9] Center for Child Development, Exercise and Physical Literacy, University Children's HospitalUniversity Medical Center, Utrecht, The Netherlands

[10] Department of Pediatric Hematology, Emma Children's Hospital, Amsterdam UMCUniversity of Amsterdam, Amsterdam, The Netherlands

Correspondence
Olav Versloot, Van Creveldkliniek (C01.428), PO Box 85500, 3508 GA Utrecht, The Netherlands.
Email: o.versloot@umcutrecht.nl

Abstract

Introduction: Although sports participation is advocated in people with haemophilia (PWH), detailed data concerning sports participation in Dutch PWH is lacking.

Aim: to assess sports participation in Dutch PWH (6-65 years) compared to the Dutch general population (GP).

Methods: Data from a nationwide, cross-sectional study in PWH were analysed. Sports participation (type, duration, frequency) was assessed by the Modifiable Activities Questionnaire (MAQ), limitations in activities using the (Paediatric) Haemophilia Activities List ((Ped)HAL). Sports in the two highest categories according to the National Hemophilia Foundation classification were considered high-risk sports. Groups were compared using Chi-square testing.

Results: A total of 524 Adult PWH (median age: 45 (IQR: 30–55); 37% severe) and 126 paediatric PWH (median age: 11 (IQR: 8–14); 52% severe) were included. Sports participation was higher in adults (70%) than the GP (58%) and similar to the GP in children (PWH: 68%, GP: 72%). High-risk sports participation decreased with age in PWH: from 65% (6-12 years) to 17% (50-65 years), which was also observed in the GP. Sports participation in children was independent of severity (non-severe: 67% vs. severe: 65%; $P = 0.97$), but not in adults (non-severe: 75%, severe: 62%; $P < 0.01$). Non-severe PWH played more high-risk sports than severe PWH: children at 65% vs. 48% ($P = 0.05$), adults at 25% vs. 15% ($P = 0.07$).

Discussion: These results suggest that sports participation in PWH was comparable to the GP. Sports participation was dependent of haemophilia severity in adults. Children were more involved in high-risk sports than adults. More studies on sports-related injury-risk are needed for adequate counselling.

KEYWORDS
children, haemophilia, injuries, joint bleeds, sports

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1 | INTRODUCTION

Haemophilia is an inherited haematological condition, causing clotting factor VIII (FVIII) of IX (FIX) deficiency.[1] Patients with low levels of clotting factor experience spontaneous or traumatic bleeds, particularly in joints and muscles leading to haemophilic arthropathy.[2] Until recently, treatment consisted of regular intravenous replacement of the missing clotting factor (prophylaxis). Novel non-replacement therapies have recently been introduced.[3,4]

Physical activity is part of a healthy lifestyle and promotes general well-being, and is recommended by the World Health Organization (WHO).[5] Sports are a fundamental element of physical activity. Among other domains (strength, balance and flexibility), the WHO recommends at least 60 min of moderate to vigorous activity per day in children and 150–300 min per week for adults to maintain or improve general health. Besides this, the WHO recommends strength (≥2 days/wk) and balance (≥3 days/wk) exercises for adults.[5] In addition, regular physical exercise is especially recommended for patients with haemophilia (PWH) to increase muscle strength and proprioception,[6] and potentially reduce bleeding risk.[7] Traditionally, PWH were advised to only participate in low-impact sports like swimming and cycling[8] and to avoid (contact) sports like soccer or basketball. The introduction of prophylaxis[9] has drastically improved the opportunities to participate in sports for PWH, making sports participation a patient relevant outcome.

Most studies on sports participation in PWH have reported on children,[10–13] data of adult PWH are limited.[14] The majority of these studies have reported on the proportion of PWH engaged in sports and type of sports,[15] without considering self-reported limitations in activities.

The primary aim of this current study was to describe sports participation in Dutch PWH according to age and severity and compared to the general population (GP), including an assessment of high-risk sports according to age and severity. A secondary aim was to assess, the association between sports participation and self-reported limitations in activities.

Bullet Points

What is already known:

- Sports participation is an important aspect of social participation
- Detailed data on sports participation in Dutch adults with haemophilia is lacking.

What this study adds:

- Sports participation among 760 adults and children with haemophilia was high and similar to the general population;
- As in the general population, older patients with haemophilia were less involved in sports.
- Sports participation was negatively associated with haemophilia severity in adults with haemophilia, but not in children.
- Children with haemophilia were more likely to participate in high-risk sports than adults.

2 | METHODS

2.1 | Design and setting

This study was part of the 6th nationwide cross-sectional 'Haemophilia in the Netherlands' (HiN6) study.[16] The HiN6 survey aimed to include all Dutch PWH or their parents. The data of the HiN6 survey were collected between May 2018 and August 2019. Participants were asked to complete a series of questionnaires covering multiple aspects of haemophilia (disease characteristics, treatment, employment, limitations, etc.). All questionnaires were either completed online or on paper. Sports participation was one aspect that was addressed in the HiN6 study. The study was approved by the Institutional Review Board of the Leiden University Medical Centre (NL59114.058.17). Participants (or their parents in those under 16) who completed the questionnaire were considered as having consented to participate (opt-in inclusion).

2.2 | Participants

Eligible patients were contacted by their haemophilia treatment centre to consider participation. Male patients with haemophilia A or B with endogenous clotting factor activity levels < 0.40 IU/mL who are registered at one the haemophilia treatment centres in the Netherlands were eligible. The present analysis included all PWH aged between 6 and 65 who completed the sports questionnaires of the HiN6 study. This age group was selected because persons between the age of 6 and 65 are most likely to be participating in organized sports in the Netherlands.

2.3 | Data collected

Data were collected on patient characteristics (age, height, weight, comorbidities), diagnosis (type, severity, comorbidities) and treatment (use of prophylaxis, treatment frequency, dose). Clinical characteristics were collected from electronic patient records, to improve reliability over self-reported data. The HiN6 included separate questionnaire-sets for children (younger than 12), adolescents (12-17) and adults (18 and older). The questionnaires for children were completed by their parents.

Patients completed validated questionnaires on sports participation (Modifiable Activities Questionnaire [MAQ][17]), physical performance (HEP-Test-Q)[18] and limitations in activities (Haemophilia Activities List [HAL][19] or Paediatric Haemophilia Activities List [PedHAL].[20]

TABLE 1 Overview of used questionnaires with domains, content, validated age categories and outcome

	Year of publication	Domains	Number of questions	Validated age categories	Outcome
MAQ	1993	- Leisure time - Professional and domestic activities	11	12-16[39]; adults[23]	Free text, energy expenditure
HAL	2004	- Lying, sitting, kneeling, standing - Functions of the legs - Functions of the arms - use of transportation - Self-care - Household tasks - Leisure activities and sports - Adaptations and using an aid	49	18-76[40]	Standardized score (0-100), higher scores indicate fewer limitations
PedHAL	2010	- Sitting, kneeling, standing - Functions of the legs - Functions of the arms - Use of transportation - Self-care - Household tasks, - Leisure activities and sports - Adaptations and using an aid	53	<8,[20] 8–17[20] (< 8: completed by parents)	Standardized score (0-100), higher scores indicate fewer limitations
HEP-Test-Q	2010	- Physical status - Mobility - Strength & coordination - Endurance - Body perception	25	6-17[41]; 24–64[18]	Standardized score (0-100), high scores indicate better subjective physical performance

Table 1 shows detailed information about the questionnaires used in this analysis.

Sports participation was defined as being actively engaged in sports at least 10 times in the last 12 months[17] (as defined in the EU Sports Charter[21]). Daily physical activities as walking or cycling to work or school or PE classes were not considered.

The MAQ[19] was validated in Dutch[23] and assesses type of sports and weekly sports exposure (frequency x duration) using free-text. Outcome consists of type of sports and weekly exposure, which can be used to calculate energy expenditure.

The HAL (49 questions) and PedHAL (53 questions) assess limitations in activities, and yields a total score (HALsum). HALsum scores are normalized to 0–100, with higher scores indicating fewer limitations.

The HEP-Test-Q[18] questionnaire assesses physical state, mobility, strength & coordination, endurance and body perception. The HEP-Test-Q was not analysed, but only used to classify participants who failed to complete the MAQ but did complete the HEP-Test-Q as sporting or not playing sports.

Sports injury risk was categorized according to the classification of the American National Hemophilia Foundation (NHF; see supplemental tables S1, S2 and S3).[24] These categories run from 1 (low risk; e.g.: swimming) via 1.5 (low to moderate risk; e.g.: rowing), 2 (moderate risk; e.g.: tennis), 2.5 (moderate to high risk; e.g.: soccer) to 3 (high risk; e.g.: field hockey). Sports in category 2.5 and category 3 were considered high-risk sports.[24] In case of a reported range in the NHF categorization (e.g.: baseball: 1.5-2.5), the median value was used.

2.4 | General population data

Sports participation data from the male GP were collected by the Dutch central bureau of statistics (CBS) and downloaded on October 17, 2018. These data were collected by means of a self-devised questionnaire by the CBS. The questionnaire was applied either online or on paper. Participants were asked in which sports they participated, including weekly frequency and duration. Both the MAQ and the CBS questionnaire included a question asking which sports were performed (answered in free text) and had a recall period of 1 month.

2.5 | Statistics

Limitations in activities were categorized into 'no limitations' (HAL > 89 or pedHAL > 95) and 'with limitations' (HAL≤89 or pedHAL≤95), based on the smallest detectable change in the HAL of 10.9 points.[25] For the present analysis, the results of children (6-11 years of age) and adolescents (12-17) and were combined to one group ("children").

Sports participation was compared according to haemophilia severity (severe, moderate, mild), age categories (6-12, 13–18, 19–29, 30–49, 50–65 years) and self-reported limitations (absent (PedHAL > 95[20] or HAL > 89[25]) or present). For the analysis of the association of high-risk sports, age and self-reported limitations in activities, patients were classified according to the highest reported sports-risk category.

Differences between sports participation and severity and age subgroups were assessed by Pearson's Chi Square testing.

All results were presented as median values with interquartile range (P25-P75, IQR) and /or proportions with 95% confidence intervals (CI) where appropriate.

Statistical significance levels were set at 5% ($P < 0.05$). The statistical analysis was performed using SPSS statistical software, version 25 (IBM corp., Armonk, NY).

3 | RESULTS

3.1 | Participants

The HiN6 study had an overall response rate of 46% (1009/2191), including 771 participants aged 6–65. The sports questionnaires (MAQ and HEP-Test-Q) were completed by 650 participants (524 adults, 126 children).

Patient characteristics for adults and children are presented in Table 2. Median age for adults was 47 years (IQR: 31–56) and 11 (8-14) for children. Haemophilia A was most prevalent in both adults and children (87%). In adults, 37% had severe haemophilia and in children 52% had severe haemophilia. Prophylaxis, consisting of infusions mostly 1–3 times/wk, was used by 164 (86%) adults and 62 (94%) children with severe haemophilia. BMI was classified as underweight (< 18,5 kg/m^2), normal weight (18,5-25), overweight (25-30) or obese (> 30) for adults. For children, BMI was classified according to age using normal values for Dutch boys. The prevalence of being overweight was similar to the GP in both adults (51% vs. 50.1%) and children (16.8% vs. 16.4%).[26,27] Adults reported more limitations in activities (median HALsum adults: 95.7 (74-100), 49% below 89) than children (99.6 (95.4-100), 21% below 95).

3.2 | Sports participation

3.2.1 | Participation – general

The MAQ was completed by 408 adults (78%) and 126 children (100%). In addition, 116 adults completed the HEP-Test-Q but not the MAQ and were included and classified as non-sporting, resulting in a total of 524 adults and 126 children available for analysis. Table 3 reports detailed sports participation for children and adults. Compared to adults, children reported a similar sports participation (68% (CI: 59–76) vs. 70% (66-74); $P = 0.66$) and significantly more HR-sports participation (55% (CI: 46–64) vs. 22% (CI: 19–26); $P < 0.01$). The most reported risk category in children was 2.5 (moderate to high risk). This was mainly due to high involvement in soccer (60%). Adults predominantly reported category 1 (low risk), especially swimming (28%).

Children reported a median weekly frequency of 4.2 (IQR: 2.1-6.5) times and a weekly exposure of 4.2 (2.7-6.7) hours of sports. Adults reported a median weekly frequency of 3.0 (1.4-5.6) times and 3.3 (1.9-6.0) hours of sports.

3.2.2 | Participation compared to the general population

Sports participation was higher than the GP in adults (PWH: 70% vs. GP: 58%; $P < 0,01$) and similar to the GP in children (68% vs. 72%; $p = 0.33$).[28] Table 4 shows differences in favourite sports between both adults and children with haemophilia and the GP. Soccer was most popular in both children with haemophilia and the GP (see supplemental table S3 for all reported sports), but the subsequent sports in the top-5 were different.

3.2.3 | Participation according to age

Sports participation in PWH was compared to the GP in age groups (6-11; 12–17;18-29;30-49; 50–65). This selection was made to enable the comparison with GP data as these data were analysed according to these groups. Figure 1 shows that sports participation was relatively stable in PWH around 70% for all age groups, although some fluctuation can be observed, particularly during adolescence and young adulthood. Sports participation in the GP showed an age-related decline in adults (18-29: 67% to 50–65: 48%). For high-risk sports however, participation showed a sharp age-related decline in both children (from 65% in 6–12 years to 40% in 13–17 years; Chi Square: $P = 0.01$) and adults with haemophilia (from 35% in 18–29 years to 19% in 30–49 and 17% in 50–65 years; $P < 0.01$). This is most likely due to the age-related decrease in soccer, which was the most popular sport in children but was rarely reported by adults (Table 4).

3.2.4 | Participation according to haemophilia severity and age

Sports participation according to age and severity is shown in table 3. Sports participation was not associated with severity in children (severe: 65% moderate: 67% and mild 67%; Chi square: p = 0.97) haemophilia. In contrast, sports participation was associated with severity in adults with haemophilia: patients with severe haemophilia (62%), were less involved in sports than those with mild (75%; $P < 0.01$) and moderate (77%; $P < 0.01$) haemophilia. High-risk sports participation was lower in those with severe haemophilia for both children (severe: 48% vs. mild: 65%; $P = 0.05$) and adults (15% vs. 25%; $p = 0.06$).

Weekly frequency was independent of severity in children (mild: median 3.7 (IQR: 2.8-5.9) vs. severe: 4.5 (1.9-7.3); $P = 0.35$) and adults (3.7 (1.9-6.1) vs. 2.7 (1.2-5.7) times/wk; $P = 0.55$). Total weekly duration

TABLE 2 Patient and treatment characteristics (n = 650)

	Adults				Children			
	Overall	Severe	Moderate	Mild	Overall	Severe	Moderate	Mild
Median (IQR) or number (%)								
number of participants	524	192 (37%)	64 (12%)	264 (50%)	126	66 (52%)	12 (10%)	48 (38%)
Age (y)	47 (31-56)	44 (30-54)	39 (28-53)	48 (34-57)	11 (8-14)	12 (8-14)	9 (8-13)	10 (9-13)
Disease and treatment (n = 650)								
Haemophilia A	455 (87%)	168 (88%)	55 (86%)	229 (87%)	107 (87%)	53 (82%)	12 (100%)	41 (85%)
Full time prophylaxis	177 (35%)	164 (86)	7 (11%)	6 (2%)	69 (55%)	62/66 (94%)	5 (42%)	2 (4%)
Prophylaxis dose (IU/kg/infusion)	14.3(11.9-22.0)	14.5(11.9-22.1)	11.4(4.8-19.7)	12.0(0.0-20.5)	24.1(17.7-32.1)	23.5(17.5-32.5)	27.8(21.1-34.7)	–
Prophylaxis Frequency (times/week)	3 (2-3.5)	3 (2-4)	3 (1 -3)	–	3 (2-3)	3 (2-3)	3 (2-3)	–
Body composition (n = 648)								
Height (cm)	183(178-187)	182(178-186)	183(179-189)	183(179-188)	150(140-170)	153(140-172)	143(133-172)	150(140-168)
Weight (kg)	85 (77-95)	83 (74-92)	87 (80-94)	86 (77-95)	40 (30.9-56.6)	42 (32-60)	40 (27-62)	40 (31-54)
% overweight	51%	46%	51%	55%	16.8%	10%	5%	6%
Comorbidities (n = 650)								
HCV ever	170 (32%)	108 (56%)	20 (31%)	42 (16%)	–	–	–	–
HIV present	17 (3.2%)	17	0	0	–	–	–	–
Functional scores (n = 634)								
Self-reported limitations (HAL; 0–100)	95.7(74.4-100)	74.5(49.5-92.8)	95.3(82.9-99.6)	99.5(94.4-100)	99.6(95-100)	99.6(95.3-100)	99.6(87.0-100)	99.4(96.6-100)
HAL < 89/PedHAL < 95	257 (49%)	151 (80%)	33 (52%)	72 (27%)	26 (21%)	14 (21%)	4 (33.3%)	8 (17%)

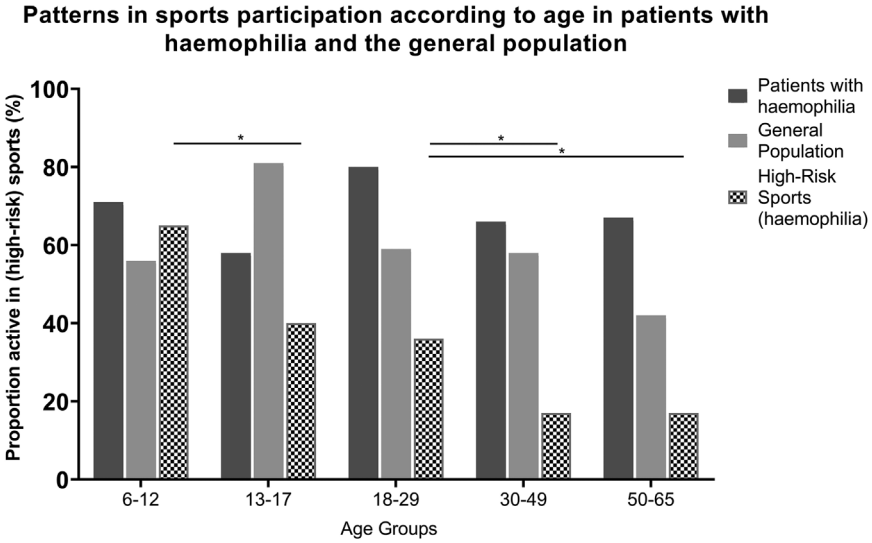

FIGURE 1 Dutch PWH show consistent sports participation and an age-related decrease in high-risk sports. Adults in the GP showed an age-related decline in sports participation. Dark grey columns represent PWH, grey columns represent the male Dutch GP (data source: gezondheidsmonitor [health monitor]).[42] Dotted columns represent high-risk sports participation in PWH, as the proportion of sporting participants that plays high-risk sports. Sports participation was defined as being actively engaged in sports (as defined in the EU Sports Charter[21]) at least 10 times in the last 12 months.[17] *: Participation in high-risk sports declined with age ($P < 0,01$)

was independent of severity in children (3.7 (2.1-7.0) vs. 3.7 (1.9-6.0) h/wk; $P = 0.30$) and adults (3.8 (1.9-7.0) vs. 2.9 (1.4-5.6) h/wk; $P = 0.86$) as well.

3.2.5 | Self-reported limitations and sports participation

Children playing sports reported similar limitations in activities as those not playing sports in the overall score (PedHALsum: median 100 (97-100) vs. 100 (91-100); $P = 0.44$). Likewise, PedHALsum scores were similar between children playing high risk sports and those playing other sports.

For adults however, those playing sports reported significantly fewer limitations in overall activities than those who did not (HALsum: 97 (83-100) vs. 66 (43-96); $P < 0.01$).

For those playing sports, the association between type of sports and self-reported limitations in activities was small. Those playing high risk sports had slightly higher HAL scores than those playing other sports: HALsum score was 99 (93-100) vs. 95 (75-100) ($P < 0.01$). However, although this difference was statistically significant, the difference was below the smallest detectable change for the HAL (11 points), suggesting limited clinical relevance.

4 | DISCUSSION

4.1 | Principle findings

This first nationwide study on sports participation in Dutch PWH showed that sports participation across all age groups was high and similar to the GP. Both children and adults with haemophilia reported high sports participation, which was relatively stable from the age of 13 years onwards. Children reported fewer limitations than adults, while adults with haemophilia who were active in sports and high-risk sports reported fewer limitations than those not involved in sports. This may be a cause or a consequence. Adults playing high-risk sports reported fewer limitations than those not playing high-risk sports. Prospective, longitudinal follow-up studies are necessary to elucidate this association.

4.2 | Internal and external validity

The overall response rate of the entire HiN6 study was 46%, but it is unlikely that (non-) participation was associated with sports participation and the study size is still considerable. A comparison between responders and non-responders was not possible as no data from the non-responders was collected. The MAQ was not completed by all participants. To reduce bias and overestimation of actual sports participation, participants who completed the HEP-Test-Q but not the MAQ were considered as not playing sports. Although we acknowledge that the use of two questionnaires is an indirect method of identifying patients who did not play sports, we believe this method to be superior to classifying non-responders based solely on the MAQ response. Without using this method, the percentage of sporting adult participants would be 89% (367/408) instead of 70% (367/524). This is much higher than the Dutch GP, suggesting selection bias. By including the HEP-Test-Q data, we were able to reduce selection bias and overestimation. Patient and treatment characteristics in the analysed data and the entire dataset were similar, giving no indication for the presence of selection bias.

Sports participation and self-reported limitations were assessed with standardized questionnaires (MAQ[17] and HAL/PedHAL[19,20], respectively) which have been extensively used in haemophilia research.[10,11,29,30] Both the data collected in the HiN6 and the GP data were self-reported, assessing sports participation over recent months.[28] Self-reported assessments often lead to an overestimation of sports participation. Particularly duration of sports is notoriously difficult to estimate for participants, leading to overestimation

TABLE 3 Sports participation according to age and severity

		children				adults			GP	
	overall	severe	moderate	mild	overall	severe	moderate	mild	Children	Adults
	Number (% [95%CI]) or median (IQR)									
Participants	126	65	12	49	524	192	64	264	2587	9978
Playing sports	68% (59-76)	65% (52-75)	67% (39-86)	67% (52-75)	70% (66-74)	62% (55-69)*	77% (65-85)	75% (69-79)	68%	54%
Playing high risk sports	55% (46-64)	48% (36-60)	58% (32-81)	65% (50-77)	22% (19-36)	15% (11-21)	31% (21-43)	25% (21-31)	-[a]	-[a]
Frequency (times/week)	4.2 (2.1-6.5)	4.6 (1.9-6.7)	2.1 (1.6-6.5)	3.7 (2.8-5.6)	3 (1.4-5.6)	3 (1.4-5.3)	3 (1.9-5.8)	3.3 (1.4-5.8)	-[a]	-[a]
Exposure (hours/wk)	4.2 (2.7-6.7)	3.7 (2.6-6.1)	2.5 (1.6-5.6)	4.3 (2.9-9.4)	3.3 (1.9-6.5)	3.4 (1.6-6.2)	3.2 (2.1-7)	3.2 (1.9-6.4)	-[a]	-[a]

* sports participation was lower in adult patients with severe haemophilia compared to non-severe haemophilia.
[a] not assessed in the questionnaire used by the Dutch central bureau of statistics.

TABLE 4 Top 5 most popular sports in children and adults with haemophilia, compared to the male general population

Children (< 12)		Children (12-17)		Adults (18+)	
PWH	GP	PWH	GP	PWH	GP
Soccer (21%)	Soccer (41%)	Soccer (15%)	Soccer (34%)	Cycling (22%)	Fitness (26%)
Swimming (19%)	Tennis (7%)	Running (8%)	Fitness (14%)	Fitness (20%)	Running (14%)
Cycling (7%)	Judo (7%)	Cycling (7%)	Hockey (6%)	Walking (14%)	Soccer (9%)
Gymnastics (7%)	Swimming (4%)	Fitness (7%)	Running (4%)	Running (9%)	Swimming (6%)
Judo (4%)	Hockey (4%)	Swimming (5%)	Tennis (4%)	Swimming (7%)	Tennis (5%)

of sports participation and physical activity.[31,32] Prospectively collecting physical activity data with an objective tool, such as an accelerometer,[33] is expected to result in more reliable study results. In this study, we have limited ourselves to sports participation, rather than physical activity in general because of the ongoing debate about the benefits and limitations of sports participation for people with haemophilia among patients, caregivers and healthcare professionals.

4.3 | Comparison with other studies

This was the first study that assessed sports participation in both children and adults with haemophilia on a national level over the same time period. In addition, the number of respondents (n = 760) is much higher than in previous studies, allowing for more detailed analyses with smaller error margins.

For adult persons with haemophilia, previous studies are scarce. Von Mackensen et al. (2016) reported high sports participation (64%; 2 times/wk, 4 h/wk) in adult PWH (n = 50; age: 35–44; 56% severe).[34] In contrast with the present study, more participants with severe haemophilia than those with mild haemophilia were active in sports (78.6 vs. 45.5%). The results of the current study were corroborated by studies in Dutch, Swedish and Irish adults with haemophilia.[14,29,35] All these studies assessed sports participation using questionnaires (self-devised, MAQ, International Physical Activity Questionnaire). These studies reported similar high sports participation (57-100%). Sherlock et al. reported 66% of Irish PWH (n = 46; age: 16–63; 49% severe haemophilia) participating in sports, with an age-related decline in sports participation. As in the current study, this study reported less sports participation in adults with severe haemophilia than in mild haemophilia and a negative association with limitations in acitivities.[35]

For children with haemophilia, most studies focused on sports injuries,[13,36–38] rather than participation. A British cross-sectional study was the only study collecting similar data to this current study (n = 84, age: 6–18). As in the current study, this study reported high sports participation with 90.5% playing sports (2x/wk; 4.9 h/wk), without comparison with the GP.[38] A retrospective study in Dutch boys with haemophilia (n = 102; age: 6–18) reported similar sports participation to the GP in (77%; 3x/wk).[13] As in this study, the MAQ was used in studies with Dutch and Australian boys with haemophilia. Both studies reported higher weekly sports exposure in children with haemophilia (8.6 and 7.9 h/wk, respectively) than the current study (4.2 h/wk). However, the groups in these studies were smaller (n = 36 (age: 8–18; 45% severe; cross-sectional) and n = 104 (4-18; 82.7% severe), respectively) than the current study.[10,11] Besides a patient-reported measure for physical activity, the Australian study. prospectively recorded activity and injuries during a one year follow-up as well.

4.4 | Perspective and clinical inference

Sports participation as well as other physically demanding activities (e.g.: labour) are an interesting and novel research area in PWH as it represents both physical status and societal participation. Although sports participation was high across the age groups, differences still exist. The source for the differences between adults and children is unknown. Potential sources are bleeding and treatment history, poorer joint status in adults and/or a more restrictive policy towards sports participation in the earlier years of older PWH. Increased sports participation comes with a concomitant increase in injury risk. Therefore, studies need to collect injury data as well. Future research should focus on life span prospective studies assessing both sports participation and sports injuries, and compare this with GP data for correct interpretation.

5 | CONCLUSION

The results of this study suggested high sports participation that was consistent with age in both children and adults with haemophilia, which was similar to the GP in both groups, although the GP showed an age-related decrease in sports participation. Adults with severe haemophilia were less involved in sports than those with non-severe haemophilia. This was not reported in children. Patients active in (high-risk) sports reported fewer limitations than those not involved in (high-risk) sports. This may be cause or consequence. The objective of haemophilia treatment is to guide patients towards a healthy (older) adulthood. In this respect, adequate counselling with regards to physical activity and sports is an important aspect of clinical care. Therefore, prospective information on type and intensity of sports performed as well as on injuries remains a necessity.

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CONFLICT OF INTEREST

OV has received speaker's fees from NovoNordisk and received research support from Bayer. All fees are paid to the institution.

EvB has not declared any conflicts of interest

SH has not declared any conflicts of interest

SS has been paid for participation in an advisory board of NovoNordisk and she received a grant to participate in a masterclass of Takeda, both in 2020.

FL received unrestricted research grants from CSL Behring, Shire/Takeda and uniQure. He is a consultant for CSL Behring, Takeda, Biomarin and uniQure, of which the fees go to the University. He received travel support from SOBI. He is DSMB member of a study sponsored by Roche.

JE has received research support from CSL Behring and honorarium for educational activities of Roche and Celgene (all paid to the institute).

MC has received financial support for research from Bayer, CSL Behring, Daiichi Saknyo, Portola/Alexion, Roche, Sanquin Blood

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LvV received a research grant from CSL Behring, is consultant for Sobi and Tremeau.

All fees are paid to the institution.

CS has not declared any conflicts of interest.

MD has not declared any conflicts of interest.

JvdN has not declared any conflicts of interest.

SG has received unrestricted research grants from Sobi.

KF has received speaker's fees from Bayer, Baxter/Shire, Biotest, CSL Behring, Octapharma, Pfizer, NovoNordisk; performed consultancy for Baxter/Shire, Biogen, CSL-Behring, Freeline, NovoNordisk, Pfizer, Roche and SOBI; and has received research support from Bayer, Pfizer, Baxter/Shire, and Novo Nordisk.

AUTHOR CONTRIBUTIONS

All authors were involved in the design of the study. E.C. van Balen and S. Hassan were involved in data collection and cleaning. O. Versloot, J. van der Net and K. Fischer analysed the data and wrote the initial version of the report. All authors were involved in data interpretation and review of the paper. All authors approved the final version of the paper.

DATA AVAILABILITY STATEMENT

The data are available to external parties upon reasonable request.

ORCID

Olav Versloot https://orcid.org/0000-0003-0748-1364
Erna C. van Balen https://orcid.org/0000-0002-3678-6581
Shermarke Hassan https://orcid.org/0000-0002-5045-636X
Frank W.G. Leebeek https://orcid.org/0000-0001-5677-1371
Lize F.D. van Vulpen https://orcid.org/0000-0003-3242-5524
Kathelijn Fischer https://orcid.org/0000-0001-7126-6613

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SUPPORTING INFORMATION

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

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