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ORIGINAL ARTICLE

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Can motor proficiency testing predict sports injuries and sports-induced bleeds in people with haemophilia?

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Abstract

Introduction: Predicting the risk of sports injuries and sports-induced bleeds (SIBs) in people with haemophilia (PWH) may support clinical counselling.

Aim: To assess the association between motor proficiency testing and sports injuries and SIBs and to identify a specific set of tests for predicting injury risk in PWH.

Methods: In a single centre, prospective study male PWH aged 6–49 playing sports ≥1x/week were tested for running speed and agility, balance, strength and endurance. Test results below −2Z were considered poor. Sports injuries and SIBs were collected for 12 months while 7 days of physical activity (PA) for each season was registered with accelerometers. Injury risk was analysed according to test results and type of physical activity (%time walking, cycling, running). Predictive values for sports injuries and SIBs were determined.

Results: Data from 125 PWH (mean [± SD] age: 25 [± 12], 90% haemophilia A; 48% severe, 95% on prophylaxis, median factor level: 2.5 [IQR 0–15]IU/dl) were included. Few participants ($n = 19$, 15%) had poor scores. Eighty-seven sports injuries and 26 SIBs were reported. Poor scoring participants reported 11/87 sports injuries and 5/26 SIBs. The current tests were poor predictors of sports injuries (Range PPV: 0%–40%), or SIBs (PPV: 0%–20%). PA type was not associated with season (activity seasonal p values > .20) and type of PA was not associated with sports injuries or SIBs (Spearman's rho < .15).

Conclusion: These motor proficiency- and endurance tests were unable to predict sports injuries or SIBs in PWH, potentially due to few PWH with poor results and low numbers of sports injuries and SIBs.

KEYWORDS
children, haemophilia, motor proficiency, sports

1 | INTRODUCTION

Haemophilia is an inherited haematological condition with impaired coagulation caused by a lack of clotting factor VIII (haemophilia A) or IX (haemophilia B).[1] People with haemophilia (PWH) specifically suffer from bleeding in muscles and joints, leading to joint destruction in the absence of proper treatment.[2] Current treatment consists of regularly self-infusing clotting factor concentrate to prevent and/or treat bleeding, although non-replacement therapy has recently been introduced.[3]

Regular physical activity is promoted by the World Health Organization for both healthy people and people with chronic conditions, such as haemophilia.[4,5] Despite established health benefits, sports participation in PWH has been discouraged for decades to minimize bleeding risk. Recently, awareness about the importance of exercise on joint stability and prevention of joint bleeds has increased. Nowadays, PWH under adequate prophylactic treatment have become as active in sports as the general population, including contact sports such as soccer or field hockey.[6,7] Independent of haemophilia, an increased sports participation leads to a higher injury risk,[8,9] with a risk of SIB being of particular concern in PWH as well as the long term effects of (joint) bleeds.[2] In order to prevent sports injuries and SIBs, identifying risk factors is clinically important. PWH may benefit from a sports injury prevention approach including prophylaxis and careful examination of the patients' physical fitness and physical activity levels.[10]

A proposed model of injury causation for sports injuries specific for PWH (including joint status, adherence and factor levels at time of injury) is shown in Figure 1.[11,12] This model includes modifiable internal and external risk factors. Especially internal risk factors may be modified by haemophilia treatment and/or specific preventive training. Most existing test protocols and prevention programmes (such as the FIFA 11+ in soccer) focus on reducing the internal risk factors for sports injuries by improving balance, strength, power, range of motion (ROM) and endurance.[9,13–20] However, such sports injury risk test protocols have not been established for PWH so far.

Before implementing a sports injury assessment protocol, its clinical relevance should be determined by assessing the predictive value of selected tests in the desired population. In a population of PWH, predicting sports-induced bleeds (SIBs) is of particular interest.

Therefore, the aim of this study was to assess the clinical relevance of motor proficiency, endurance tests and type of physical activity to predict sports injuries and SIBs in a cohort of PWH actively involved in physical activity and sports.

2 | METHODS

2.1 | Methods and setting

This study was performed as part of the 'Sports Participation and Injuries in People with Haemophilia' (SPRAIN) study, a single-centre, prospective longitudinal study and was performed at the Van Creveldkliniek of the UMC Utrecht.

The study was approved by the Internal Review Board of the UMC Utrecht (approval number: 18–141). All participants signed informed consent prior to participating in the study. In case of children, both parents or caregivers provided informed consent.

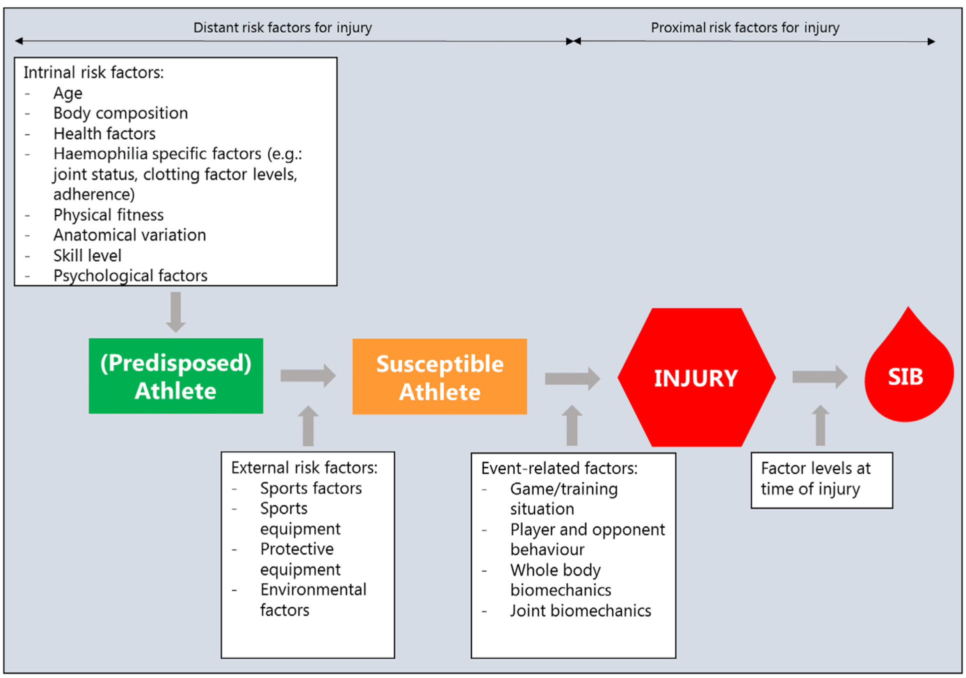

FIGURE 1 An example of a comprehensive model for injury causation showing potential internal and external risk factors for the occurrence of sports injuries and sports-induced bleeds. Adapted specifically for people with haemophilia (PWH) from Bahr and Krosshaug (2005) to include haemophilia specific risk factors.[12]

All participants were included between October 2018 and March 2020, when inclusion was discontinued prematurely due to the COVID-19 lockdown. Follow-up was 12 months. After baseline testing, participants were contacted every 2 weeks to record injuries and bleeds. The study was registered in the Dutch trial register (www.trialregister.nl) under trial ID NTR6769.

2.2 | Participants

PWH aged 6–49 at inclusion, who participated in sports at least once weekly were invited to participate in the study. The presence of FVIII/FIX antibodies, an arthrodesis or arthroplasty within the last 12 months or neurological disabilities were exclusion criteria for participation. All haemophilia severities were included.

2.3 | Data collected

Baseline testing consisted of motor proficiency and endurance tests. Motor proficiency was assessed by means of selected domains of the Bruininks–Oseretsky Test 2 (BOT-2).[21,22] The BOT-2 was developed to assess motor proficiency in children and has been validated in various populations.[23–25] It consists of eight domains: fine motor precision, fine motor integration, manual dexterity, bilateral coordination, balance, running speed & agility, upper-limb coordination and strength. A composite score can be calculated when all domains of the BOT-2 are tested. However, not all domains were considered clinically relevant to assess sports injury risk, especially in a population active in contact sports. The domains were selected based on several criteria: assessment of known risk factors for sports injuries,[12–17] a clear description in a manual and no need for additional training or the use of complex equipment. Furthermore, as the selected tests assess universal skills (strength, balance, endurance), we reasoned that these tests would be valid in adults as well and this would not negatively affect the validity of the current study.

The domains balance (single-leg stance, single-leg stance on balance beam, heel-to-toe stance on balance beam), strength (push-ups, sit-ups, wall sit) and running speed and agility (standing long jump, single leg hop, single leg side hop, two legged side hop) were selected for this study based on their expected clinical relevance. Running speed & agility was used as a proxy for lower extremity power (fast/explosive development of speed). The tests were conducted according to protocol of the BOT-2 test.[26] The other domains were excluded as these were not considered relevant for prediction of sports injuries and SIBs. PWH older than 22 were included in this study as well. However, as the analysis was limited to an intragroup comparison based on the selected domains rather than the composite score of the BOT-2, this seems unlikely to negatively affect the validity of these domains. Furthermore, the selected domains cover universal motor action, which are expected to be valid in extended age categories (6–49) as well, especially as no people in frail ages were included. The results of the tests in the various domains ('balance', 'running speed & agility' and 'strength') were grouped. If a participant had a 'poor' score on one of the individual tests of the domain, the entire domain was classified as 'poor'. Predictive performance of the motor proficiency tests was evaluated by means of the positive and negative predictive values (PPV and NPV, respectively) for the motor proficiency tests. A positive predictive value is calculated as the proportion of participants with a sports injury or SIB divided by the number of participants with poor test results. A negative predictive value is calculated as the number of participants with no sports injury or SIB divided by the number of participants with a poor test result. PPV and NPV were calculated separately for sports injuries and SIBs and for poor and average-to-good results.

Based on its performance in both children and adults with chronic conditions, endurance was assessed by the steep-ramp test.[27–31] The test was performed according to the protocol described previously for adults[30] and children.[31] The test ended when a participant was no longer able to maintain the required pedalling frequency despite strong verbal encouragements, or due to any physical complaints from the participant (nausea, dizziness, etc.). Peak work load and peak heart rate were recorded. Peak work load was defined as the resistance at the moment pedalling frequency dropped below 60 rpm. Peak heart rate was defined as the maximal attained heart rate during the test. An effort was considered to be maximal when participants showed subjective signs of intense effort (e.g., unsteady biking, sweating, facial flushing, clear unwillingness to continue despite strong verbal encouragement).[31]

Following inclusion and baseline testing, injuries and bleeds were prospectively collected during a 12-months' follow-up. Participants were contacted by the researcher every 2 weeks by their preferred method to check if any injuries had occurred during the previous 2 weeks. In case of an injury or bleed, the researcher would call the participant to record the details of the injury on a standardized form (see Table S1 for details).

A *sports injury* was defined as 'any injury as a result of participation in sport with one or more of the following consequences: (a) a reduction in the amount or level of sports activity; (b) a need for (medical) advice or treatment; or (c) adverse social or economic effects'.[32]

Bleeds were classified according to the ISTH definition.[33] Definitions are shown in Table S2. A bleed was considered a SIB when the bleed occurred as a result of a sports injury, had identical symptoms to a bleed and required extra infusion of clotting factor concentrate or consultation with a haemophilia consultant.

This culminated in a dataset including data from the selected tests, sports injuries and SIBs. These SIBs were a subset of the reported sports injuries.

Sports injury risk was classified according to the National Hemophilia Federation (NHF) classification. The NHF used five categories to classify injury risk in sports (1: safe; 1.5: safe to moderate risk; 2: moderate risk; 2.5: moderate to high risk; 3: high risk). Sports in the highest two categories (e.g., soccer, hockey) were considered high-risk sports. Sports injury mechanisms were subdivided in 'intrinsic' (physical fitness, limited skills), 'extrinsic' (equipment, protective equipment, environment) and 'other' to enable analysing the source of the injury or SIB.[12]

In order to objectively support the physical activity data collected with the MAQ, physical *activity* data were collected using an Activ8 (Activ8, Valkenswaard, The Netherlands) 3-axial accelerometer (30 × 32 × 10 mm; 20 g) as well. Participants were asked to wear the accelerometer for 24 h per day for 7 consecutive days, with a minimum of 4 consecutive days containing data days to be included for analysis. Data are presented as proportion of 24 h. The Activ8 is a valid, accurate tool to assess physical activity[34–36] and has been used in a wide array of medical conditions, including PWH.[37] The Activ8 is worn on the thigh and can distinguish lying, sitting, standing, walking, running and cycling, both in time and energy expenditure. Participants were asked to wear the Activ8 continuously for 7 days. For this purpose, the Activ8 was attached to the right upper thigh with a waterproof, transparent Tegaderm self-adhesive plaster (see Figure S1). Time spent lying, sitting, standing and walking were reported in h/day, running and cycling were reported in min/day. Time spent lying, sitting and standing was classified as 'passive activities'. Walking, running and cycling were classified as 'dynamic activities'.

Following an interim analysis on complete data (4 weeks of ≥4 days of data) of 15 first participants which showed similar physical activity patterns between the various seasons for all activities as measured by the Activ8 (p values from Chi square tests for variation across season: Lying: $p = .55$; Sitting: $p = .20$; Standing: $p = .29$; Walking: $p = .85$; Cycling: $p = .63$; Running: $p = .46$), it was decided to collect data for 1 week only in subsequent participants.

2.4 | Statistical analysis

Descriptive injury data were presented as medians and interquartile ranges (IQR) and/or proportions with 95% confidence intervals (95% CI), as appropriate. Z-scores were computed for each domain of the BOT-2 test and the Steep Ramp Test. A score of Z ← 2 was considered a 'poor' score, a score of Z > 2 was considered a 'good' score. Scores between −2 and 2 were considered 'average'. Data were analysed with (non-)parametric techniques, depending on the distribution of the data. This was done for separately for children and adults. Participants with moderate and mild haemophilia were grouped as 'non-severe' due to the low number of participants with moderate haemophilia in this study.

Outcomes were compared with non-parametric Spearman, Chi Square and Mann-Whitney tests, as appropriate. A p-value < .05 was considered statistically significant. Sports participation changed as a result of COVID-19 restrictions in the Netherlands, especially from high-risk team sports (e.g., soccer) to low-risk individual sports (running and walking).[38] Therefore, an additional analysis was performed in which the number of sports injuries and SIBs as well as predictive test results before and during COVID-19 restrictions were compared. An additional analysis was performed to assess the predictive values in soccer, as this was the most frequently reported sports.

The online electronic data capturing tool CASTOR was used to collect, record and store patient characteristics, injury data and test results.[39] The statistical analysis was performed using SPSS statistical software, version 26 (IBM corp., Armonk, NY, USA).

2.5 | Data sharing statement

External parties can have access to the data upon reasonable written request to the investigators.

3 | RESULTS

3.1 | Participants

Test results, injury- and bleeding data from 125 participants (41 children [median age: 13 (IQR 10–16)], 84 adults [30 (IQR 23–39)]) were collected for this study. Table 1 shows participant and treatment characteristics. Most participants had haemophilia A (90%) and almost half had severe haemophilia (47%). Nearly all participants with severe haemophilia (95%) received prophylactic replacement therapy at a median weekly treatment dose of 42 (A: 42; B: 52) IU/kg. All participants had a complete, 12 months' follow-up. Sports participation decreased during the COVID-19 pandemic, and shifted from high-risk team sports (e.g., soccer) to low-risk individual sports (e.g., running and fitness).[38] Only a minority (39/125; 31%) contacted the haemophilia treatment centre in case of a suspected sports injury or bleed.

3.2 | Sports participation, sports injuries and sports-induced bleeds

All participants were involved in sports at least once weekly (median annual exposure: 146 (IQR 65–227) h), an overview of all reported sports is shown in Table S3. Participants were engaged in 43 sports, including 74/125 (59%) in high-risk sports (see Figure S2 for the distribution of NHF risk categories). Adults were mostly engaged in fitness (29%), running (16%) and soccer (8%); children mostly played soccer (40%), fitness (7%) and gymnastics (4%). A total of 87 sports injuries were reported by 51 (41%) participants, of which 26 (20 participants; 16%) resulted in SIBs, indicating a very low risk for SIBs during sports. More participants with severe haemophilia reported injuries (53% vs. 29%; $p < 0{,}01$) and SIBs (23% vs. 9%; $p = 0{,}03$) than those with non-severe haemophilia. During COVID-19 restrictions, Participants reported more injuries (median 1 [IQR 1–2] vs. 0 [IQR 0–1]; $p < .01$) but similar SIBs (0 [IQR 0–0)] vs. 0 [IQR 0–1]; $p = .09$).

3.3 | Testing

3.3.1 | Motor proficiency levels

Figure 2 and Table S4 show the distribution of the standardised individual test results. Only a minority of the participants reported poor motor proficiency scores (below −2Z): over 90% (mean: 96% ± 1.6) of participants had average-to-good scores (all score higher than −2Z) on

TABLE 1 Participant, disease, treatment characteristics and joint health of participants with haemophilia.

N = 125	Overall	Severe	Non-severe
N (%), or median (IQR)			
Number	125	60	65
Age (years)	23.1 (15.9–33.7)	23.0 (15.8–34.1)	23.8 (16.1–33.1)
Haemophilia A	112 (90%)	54 (92%)	58 (88%)
Baseline factor activity (%IU)	2 (0–15)	0 (0–0)	14.5 (8.3–16.8)
Severe haemophilia	59 (47%)	–	–
Prophylactic treatment	61 (49%)	56 (95%)	5 (8%)
Prophylactic dose IU/kg/week	42.3 (35.8–55.1)	42.0 (35.2–55.1)	52.0 (39.1–73.7)
History of anti FIVIII/IX antibodies[a]	17 (14%)	14 (82%)	3 (18%)
Sports participation			
Annual sports exposure (h/year)	146 (65–227)	156 (77–250)	144 (64–220)
High-risk sports	74 (59%)	41 (69%)	33 (50%)
Energy Expenditure (METs-h/week)	19.4 (8.1–34.6)	16.6 (7.9–34.6)	21.7 (8.1–34.6)
High-intensity sports	25 (20%)	12 (20%)	13 (20%)
Follow-up before COVID	27 (22%)	22 (81%)	5 (19%)
Follow-up during COVID	98 (78%)	38 (39%)	60 (61%)

Abbreviation: IQR, interquartile ranges.

[a]People with haemophilia under prophylactic treatment sometimes develop antibodies against FVIII/FIX concentrates.

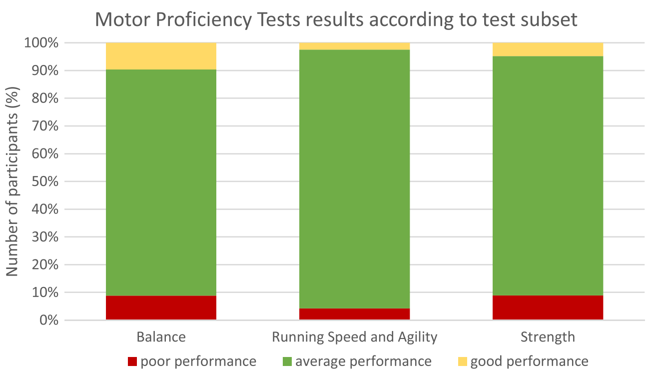

FIGURE 2 Motor proficiency test results according to BOT-2 subset. The majority of participants showed an average score on the composite test groups 'balance', 'running speed and agility' and 'strength'. Only a minority (2.5%) had a poor performance.

all tests. Standing on 1 leg with eyes open most frequently scored as 'poor' (7%).

3.3.2 | Endurance

A total of 123 participants completed the Steep Ramp Test for endurance. One participant was unable to perform the test due to limited ROM in the knee, the other had an unusually high heart rate before starting the test. This test was subsequently cancelled. Median maximal workload was similar in children and adults (children: 4.9 W/kg [IQR 4.3–5.6]; adults: 4.9 W/kg [IQR 4.3–6.0]), corresponding to an estimated median VO_2-max of 2.2 (IQR 1.5–3.0) in children and 3.0 (IQR 2.9–3.4) in adults. Only two participants (1.6%) scored 'poor' on the endurance test. These participants reported no sports injuries or SIBs.

3.3.3 | Sports injuries according to motor proficiency and endurance results

Table 2 shows the positive and negative predictive value (PPV and NPV, respectively) of the motor proficiency tests. Missing data were caused by physical impairments of the participants (limited ROM or painful joint). None of the tests could identify risk factors for sports injuries or SIBs. The PPV for sports injuries was below 50% for all domains (Balance: 36% [CI 15%–68%]; Running Speed and Agility: 40% [CI 12%–77%]; Strength: 27% [CI 9%–57%]; Endurance: 0% [CI 0%–71%]), and SIBs (Balance: 9% [CI 0%–40%]; Running Speed & Agility: 20% [CI 2%–64%]; Strength: 18% [CI 4%–49%]; Endurance: 0% [CI 0%–71%]; see Table S5). The wide confidence intervals reflect the low number of participants with low scores and the few reported sports injuries and SIBs. Table S8 shows predictive values according to severity. The PPV for both sports injuries and SIBs for severe and non-severe haemophilia were all below 50%, that is, non-informative. An additional analysis using −1SD as a cut-off value for a 'poor' result showed similar results for PPV and NPV (see Table S9). Injury mechanism was similar across PWH with poor scores (intrinsic: 8/19 vs. extrinsic: 8/19; 3/19 no injury; $p = .10$). An intrinsic source had a limited PPV (sports injury: 38%; SIB: 25%). Although PPV during COVID-restrictions were higher than before COVID, they remained below 50% for all tests with very

TABLE 2 Predictive values for sports injuries and sports-induced bleeds according to motor proficiency and endurance.

$N = 125$	n	Predictive value	
		Sports injuries	Sports-induced bleeds
Balance			
Poor	11	PPV: 36%	PPV: 9%
Average-to-good	114	NPV: 59%	NPV: 83%
Running speed and agility			
Poor	5	PPV: 40%	PPV: 20%
Average-to-good	114	NPV: 59%	NPV: 85%
Strength			
Poor	11	PPV: 27%	PPV: 18%
Average-to-good	113	NPV: 58%	NPV: 84%
Overall motor proficiency testing			
Poor	19	PPV: 37%	PPV: 16%
Average-to-good	106	NPV: 58%	NPV: 84%
Endurance			
Poor	2	PPV: 0%	PPV: 0%
Average-to-good	120	NPV: 59%	NPV: 84%

Note: PPV: positive predictive value, the number of participants who sustained a sports injury or sports-induced bleed with a poor test result (e.g., four sports injuries in 11 poor results on balance tests: 4/11 = .36). NPV: negative predictive value, the number of participants who did not sustain a sports injury or sports-induced bleed after an average-to-good test result (e.g., 47 sports injuries in 114 'average-to-good' scores on balance tests: NPV = (114–47)/114 = .59).

wide confidence intervals (see Table S7). Participants active in low-risk sports (NHF 1, 1.5, 2)[40] with poor test results, reported more sports injuries than those with average-to-good results (median 1 [IQR: 1–1] vs. 0 [0—1]; $p < .01$), while SIBs showed no association with test results for both high-risk and low-risk sports.

The large variation in sports ($N = 43$, see Table S3) and the range in test results, may have contributed to the low PPV of the various tests. Because soccer was the most frequently performed sport ($n = 39$; high-risk sport), a subgroup analysis was conducted to determine the PPV and NPV for the tests in soccer players (Table S6). Despite high PPV values for the motor proficiency tests in the soccer group (Balance: 67% [CI 20%–94%]; Speed and Agility: 100% [CI 29%–105%]), the numbers of PWH with poor results and the number of reported sports injuries and SIBs were too low to provide a clinically relevant prediction, as reflected by the wide confidence intervals.

3.4 | Physical activity

One hundred and one participants (81%) had sufficient physical activity data collected with an Activ8® activity tracker. Missing data was caused by malfunctioning equipment ($n = 22$) or skin irritation from the device ($n = 2$). Figure 3 shows the overall distribution of physical activity/24 h and the distribution according to season and shows that no seasonal variation in physical activities was observed (Lying: $p = .55$; Sitting: $p = .20$; Standing: $p = .29$; Walking: $p = .85$; Cycling: $p = .63$; Running: $p = .46$).

Participants spent the majority of time sitting (51.5%) and only limited time in high-intensity activities (cycling and running combined: 1%). Overall, participants spent 90% (~22.5 h/day) in passive activities, of which median 23% (IQR 19%–29%) lying (including sleep) and only 10% in dynamic activities. Dynamic activities were similar in children and adults (median 12% [IQR: 10–16] vs. 10% [8–13]; $p = .07$), but children spent more time in high-intensity physical activity (cycling [1.6 vs. .6%; $p < .01$] and running [.3% vs. .08%; $p < .01$]) than adults.

3.5 | Injuries and SIB according to physical activity

Sports injuries and SIBs were not associated with types of physical activity. Table 3 shows that the proportion of time spent in dynamic activity was similar in participants with sports injuries and those without sports injuries (10% [6–14] vs. 11% [8–14]; $p = .21$) and those with and without SIBs (9% [6–14] vs. 11% [9–14]; $p = .25$). Neither was time spent in dynamic activities associated with the number sports injuries (Spearman's rho: .11; $p = .28$) or SIBs (rho: .04; $p = .73$).

4 | DISCUSSION

4.1 | Principal findings

This was the first study aiming to identify internal, physical risk factors (see Figure 1) for sports injuries and SIBs in PWH. The selected physical tests included tests of motor proficiency, endurance, and type of physical activity. Very few participants ($n = 19$; 15%) had poor scores and/or sports injuries ($n = 51$; 41%) or SIBs ($n = 20$; 16%). In the current dataset motor proficiency testing, endurance and physical activity were unable to identify PWH with an increased risk of sports injuries or SIBs. Furthermore, the results indicated that types of physical activity levels showed no variation throughout the annual seasons.

4.2 | Strengths and limitations

This was a 12-month prospective study assessing injury risk based on motor proficiency testing in PWH. As this study focused on sports injuries, only PWH active in sports were included. This makes the generalizability limited to active PWH and those aspiring to become active, which is the population at risk for sports injuries. Potential selection bias seemed limited as the majority (~70%) of Dutch PWH is active in sports.[6,7]

The tests for this study were performed between October 2018 and March 2020, with a 12-month follow-up until March 2021. The

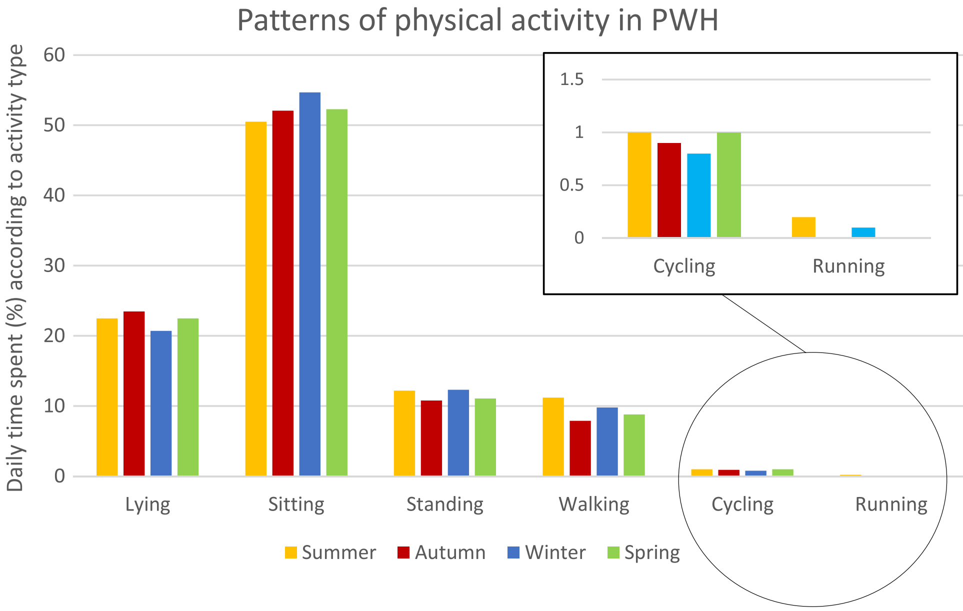

FIGURE 3 Patterns of seasonal physical activity in people with haemophilia. No seasonal variation was observed in types of physical activity.

TABLE 3 Sports injuries and sports-induced bleeds according to physical activity pattern as measured by Activ8® accelerometers.

Overall (median proportion (IQR))						
	Sports injury	**No sports injury**	***p***	**Sports-induced bleed**	**No sports-induced bleed**	***p***
Time spent in dynamic activities	10% (6%–14%)	11% (9%–14%)	.21	9% (6%–14%)	11% (9%–14%)	.25
Children (median proportion (IQR))						
Time spent in dynamic activities	11% (7%–16%)	13% (11%–16%)	.23	15% (8%–18%)	12% (9%–16%)	.40
Adults (median proportion (IQR))						
Time spent in dynamic activities	9% (6%–14%)	10% (8%–13%)	.43	8% (5%–11%)	11% (8%–14%)	.06

Abbreviation: IQR, interquartile ranges.

COVID-19 pandemic and restrictions imposed in March 2020 caused a shift in sports behaviour in Dutch PWH, particularly from high-risk team sports (NHF categories 2.5 and 3) to low-risk individual sports (NHF 1, 1.5 and 2).[38,40] This has potentially caused an underestimation of the true number of injuries, making it difficult to assess the predictive value of motor proficiency testing. However, an additional analysis showed that although participants reported more injuries but similar SIBs after COVID-19 restrictions were imposed, the PPV remained similar for all tests.

Sports injuries and SIBs were self-reported. Only 31% of suspected SIBs were evaluated by a haemophilia physician. Although specific data are lacking, this was in line with clinical practice: most patients are on home treatment and only contact the treatment centre in serious situations.

The intense monitoring of injuries and bleeds might have increased the risk of over reporting of sports injuries and SIBs, potentially leading to an actual overestimation of the PPV of the test results. For bleeding episodes, it has been established that both physicians and PWH regularly misinterpret musculoskeletal complaints as (joint) bleeds and vice versa.[41] This is in line with clinical practice, and all SIBs presented here were treated as a true bleed.

Although participants with severe haemophilia reported more injuries and bleeds than those with non-severe haemophilia, these participants reported shorter time-loss, which is indicative for less severe injuries. This could suggest that these participants have a lower threshold to report injuries and/or have developed a strategy to minimize injury disk during sport, for example, by being physically well prepared, more cautious during sports or ensuring appropriate factor levels during sports.

Although both the PPV and NPV were calculated to evaluate the test results, the PPV seems to be more clinically relevant in this case. Unfortunately, clinical inference and interpretation are hampered by the wide confidence intervals on the PPV.

This study lacked normative data from the general population regarding the outcome of the BOT-2 domains. Studies comparing results from patient populations (obesity, hypermobility syndrome, developmental disorder, sickle cell disease) and healthy children showed lower results for patients.[42–45] However, unlike the population in this study, these groups were less involved in sports/physical activity than the general population.[6,7] As motor proficiency is associated with sports participation,[46] the present study population is expected to show similar results as the general population.

4.3 | Comparison with other studies

No other studies were identified that used motor proficiency testing to predict sports injuries and SIBs in PWH. As this was a highly active population with an injury prevalence comparable to the general population,[6,7] the outcome of the current study was compared to studies assessing injury risk with different tools in the general population.

The present study used specific components of the BOT-2 (Balance, Running Speed & Agility and Strength) to assess balance, power and strength. These data could be compared to those of previous studies assessing specific physical risk factors with similar constructs as the BOT-2 domains such as the Functional Movement Screen (FMS; balance, strength),[47,48] Star Excursion Balance Test (SEBT; balance)[49] or the Drop Jump Screening Test (DJST; balance, running speed & agility).[50]

In contrast to the present study, studies in the general population identified limited balance,[17,51,52] poor strength,[53] and poor endurance[20] as risk factors for sports injuries. This discrepancy could be due to the size and heterogeneity of our population in comparison with larger, more homogeneous groups (e.g., athletes in the same sports, soldiers) in other studies. In addition to increased statistical power based on study size, these populations potentially had a higher sports exposure with a concomitant higher injury risk. Furthermore, differences in the tests used may have contributed to discrepancies in results.

Single leg stance was used as a proxy for balance. In contrast to the current study, it was identified as a risk factor in three previous studies involving young athletes and students (basketball [n = 210; mean age 16 years],[51] and Australian football [n = 210, aged 22 years][52] and students [n = 230; aged 18 years]).[17] Differences in results might be due to younger participants, higher baseline risk and higher exposure to high-risk sports (basketball, Australian football).

The systematic review identifying upper body strength as a predictor for sports injuries evaluated push-ups and sit-ups to predict injuries in adults (18–65 years). Push-ups (n = 16,968) were identified as predictors in 15/22 studies, while sit-ups (n = 16,605) were identified in 5/24 studies.[53] In both cases, study protocols were similar to the current study.

For assessment of endurance, the steep ramp test was unable to predict injuries. This is in contrast with a systematic review (49 studies; 171,318 adults) that reported an association between poor endurance and musculoskeletal injuries in 34/49 studies in soldiers and athletes. However, all these tests assessed endurance with running tests,[20] potentially explaining the different conclusions. No studies were identified in which cycling tests were used to predict injuries.

Running speed and agility test were unable to identify lower extremity power as risk factor. This was corroborated by a recent systematic review showing that nine out 11 selected studies (3257 participants; age: 18–65) reported no association between lower body power and musculoskeletal injuries.[54] Two other studies involving 71 collegiate basketball players (age 20.2 ± 1.9 years) and 127 Swedish soccer players (aged 16–36) both failed to establish an association between lower extremity power and sports injuries as well.[55,56]

4.4 | Clinical relevance and future directions

The current study was unable to identify risk factors for sports injuries and SIBs from motor proficiency test or physical activity. This might be based on low statistical power due to a small proportion of participants with poor scores who reported sports injuries or SIBs or a selection of irrelevant tests for PWH, although these tests were chosen based on previous studies in athletic populations.[9,13–19] The current tests were selected based on their practical applicability, clear description and lack of need for complex equipment. There does not seem to be an indication that including more complex measuring methods (e.g., isokinetic testing for strength testing) would lead to more precise results.[57–62] Sports injuries and SIBs are generally the result of a complex interaction between internal, external and event-related risk factors.[12] This study focused on selected internal risk factors, resulting in a limited approach from an environmental perspective. Both internal and external risk factors (e.g., sport specific risk factors, motivation, behaviour during sports and equipment) in sports should at least be considered to create a more complete overview of sports injury risk in individual PWH. The lack of seasonal variance showed that seasonal recording of physical activity did not provide an additional value in this cohort. This suggests that a single assessment is sufficient to assess physical activity, which would decrease the participant burden of future studies.

5 | CONCLUSIONS

This was the first study attempting to identify internal, physical risk factors for sports injury and SIBs in PWH. The majority of participants who reached 'average' (from −2Z to +2Z) results in the current selection of tests showed no association with sports injuries or SIBs. This is likely due to few participants with poor test results and few reported sports injuries and SIBs during the 12-month' follow-up. This study focused on internal, physical risk factors, while injury risk is dependent on internal, external and event-related risk factors. Thus, focusing on internal, physical risk factors reflects a limited approach on injury prediction. When counselling PWH regarding sports participation, a complete individualized assessment of internal (e.g., clotting factor activity levels), external (e.g., surface and (protective) equipment) and event-related factors (NHF category, playing level, behaviour) should be made to enable proper advice PWH on reducing sports injury risk.

AUTHOR CONTRIBUTIONS

O. Versloot, K. Fischer, J. van der Net, C. F. van Koppenhagen were involved in the design of the study. O. Versloot 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.

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CONFLICT OF INTEREST STATEMENT

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DATA AVAILABILITY STATEMENT

External parties can have access to the data upon reasonable request to the authors.

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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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