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Development and validation of a new questionnaire for the assessment of subjective physical performance in adult patients with haemophilia – the HEP-Test-Q

S. VON MACKENSEN,*[1] D. CZEPA,†[1] M. HERBSLEB‡ and T. HILBERG†

**Institute and Policlinics of Medical Psychology, University Medical Center Hamburg-Eppendorf, Hamburg, Germany; †Department of Sports Medicine, University of Wuppertal, Wuppertal, Germany; and ‡Department of Sports Medicine, Friedrich-Schiller-University, Jena, Germany*

Summary. Specific research studies for the investigation of physical performance in haemophilic patients are rare. However, these instruments become increasingly more important to evaluate therapeutic treatments. Within the frame of the Haemophilia & Exercise Project (HEP), a new questionnaire, namely HEP-Test-Q, has been developed for the assessment of subjective physical performance in haemophilic adults. In this article, the development and validation of the HEP-Test-Q is described. The development consisted of different phases including item collection, pilot testing and field testing. The preliminary version was pilot-tested in 24 German HEP-participants. Following evaluation and preliminary psychometric analysis, the HEP-Test-Q was revised. The final version consists of 25 items pertaining to the domains 'mobility', 'strength & coordination', 'endurance' and 'body perception', which was administered to 43 German haemophilic patients (43.8 ± 11.2 years). Psychometric analysis included reliability and validity testing. Convergent validity was tested correlating the HEP-Test-Q with SF-36, Haem-A-QoL, HAL and the Orthopaedic Joint Score. Discriminant validity tested different clinical subgroups. Patients accepted the questionnaire and found it easy to fill in. Psychometric testing revealed good values for reliability in terms of internal consistency (Cronbach's $\alpha = 0.96$) and test-retest reliability ($r = 0.90$) as well as for convergent validity correlating highly with Haem-A-QoL, HAL and SF-36. Discriminant validity testing showed significant differences for age, hepatitis A and hepatitis B and the number of target joints. HEP-Test-Q is a short and well-accepted questionnaire, assessing subjective physical performance of haemophiliacs, which might be combined with objective assessments to reveal aspects, which cannot be measured objectively, such as body perception.

Keywords: adults, exercise, haemophilia, physical performance, questionnaire, self-assessment, validation

Introduction

Recurrent musculoskeletal haemorrhages in people with haemophilia (PWH) lead to restrictions in the locomotor system. The reduced level of physical performance in patients with haemophilia affects activities of daily life in comparison with healthy subjects [1–3]. In turn, deficiency of conditional and coordinative abilities is associated with an increased frequency of joint bleeds. To stop this vicious circle, sport- and physio-therapeutic treatments are recommended [4–7].

Since 2000, the Haemophilia & Exercise Project (HEP) has been underway with the aim to improve the health status in patients with haemophilia, especially the joint function by means of specific sports therapy. Sports camps have been organised twice a year at the Regional School of Sports of Thuringia (Germany), where patients have been instructed for their later home training programme regarding 'body perception', 'muscle relaxation tech-

[1]These authors contributed equally to this study.

Correspondence: Sylvia von Mackensen, Institute and Policlinics of Medical Psychology, University Medical Center Hamburg-Eppendorf, Martinistraße 52, 20246 Hamburg, Germany.
Tel.: +49 172 682 2759; fax: +39 02 5503 2072;
e-mail: s.mackensen@uke.uni-hamburg.de

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niques', joint mobilization' as well as 'muscle activation' [8]. Details about the project can be found under http://www.haemophilia-exercise.de.

To control therapeutic treatments, specific research methods become more and more important. But, specific research studies for the assessment of physical performance in haemophilic subjects are rare [9,10].

In addition to bleeding frequency and joint function, the assessment of patients' perspective with so called patient-reported outcomes (PRO) is increasingly considered necessary to understand which treatment strategy is clinically most effective and fits the patient's needs best [11]. Patient-reported outcomes include measures assessing quality of life (QoL), treatment satisfaction, patient preferences and functioning. The development of PRO measures has advanced rapidly and implementation in clinical research and practice is now underway.

Haemophilia is a rare health condition with cost-intensive treatment strategies in which traditional outcome measures such as mortality are no longer significantly influenced by diverse treatment options and PRO assessment is increasingly recognized as important [12].

For the assessment of functional health, only one haemophilia-specific self-assessment questionnaire is available: the Haemophilia Activities List (HAL) [13]. It assesses in particular what activities of daily living the patient can do. The purpose of this study was to have a patient-reported instrument allowing the subjective assessment of different aspects of the HEP such as mobility, strength and body perception, which can be compared with the objective measurements of the HEP.

In the frame of HEP, a PRO for the assessment of subjective physical performance in adult patients with haemophilia was developed, named HEP-Test-Q. In this article, the development and validation of the HEP-Test-Q is described and its psychometric characteristics are analysed.

Materials and methods

Instrument development

The HEP-Test-Q was based on the modular training programme of the HEP. Items were chosen together with experts in sports medicine in 2005. The preliminary version consisted of 32 items, which belonged to six dimensions: 'physical status', 'mobility', 'strength & coordination', 'endurance', 'body perception' and 'general questions'. In addition, one item evaluated the physical activity compared with the past year, which was separately analysed. Respondents were asked about their physical performance over the past four weeks. The response options were a five-point Likert scale (ranging from 1 = never to 5 = always). Some of the items had to be re-coded; subscales and the total score were transformed to a scale of 0–100 with high scores indicating better physical performance.

Pilot testing

This preliminary version of the HEP-Test-Q was pilot-tested in 24 German PWHs, who participated in the HEP. The mean age of the subjects was 43.7 ± 10.7 years (range, 24–64) with a body mass index (BMI) of 26.2 ± 5.7 kg m^{-2} (range, 18–39). In all, 95.8% had haemophilia A and 87.5% were severely affected by haemophilia. Pilot testing included:

1. preliminary psychometric testing on item level (missing values, difficulty index, item-total correlation, alpha if item omitted),
2. evaluation of the questionnaire (acceptance, relevance, comprehensibility) and
3. psychometric testing on scale level in terms of reliability (internal consistency and test-retest-correlation) and validity (convergent and discriminant).

This preliminary version was subsequently revised and analysed based on the new scale with 23 items pertaining to five dimensions plus one additional item.

Field testing

Two additional items were included based on patients' recommendations; they mentioned that the items should be differentiated between (i) acute and chronic pain and (ii) walking upstairs and downstairs. The new version consisted of 25 items pertaining to the following four domains: 'mobility', 'strength & coordination', 'endurance' and 'body perception' plus one item evaluating the physical activity compared with the last year to be separately analysed. The revised questionnaire was administered to 43 German PWHs aged 43.8 ± 11.2 years (range, 19–65) with a BMI of 25.7 ± 4.9 kg m^{-2} (range, 18–39).

Additional information

In the scope of the HEP, further information was collected concerning individual-related, objective and other subjective parameters. Patients were asked

about sociodemographic data (such as marital status, educational level), clinical data (such as type of haemophilia, severity, bleeding events, treatment modalities, viral infections) and their frequency of physical activities per week.

Objective physical functioning was recorded by maximal isometric muscle strength and joint status. The maximal isometric muscle strength was measured using a knee extensor (Schnell™, Peutenhausen, Germany) in a sitting position with a knee angle of 60°. The joint status was examined using the *Orthopaedic Joint Score (OJS)* [14]. OJS is composed of the clinical score (e.g. swelling, muscle atrophy, crepitating), the pain score and the bleeding score. All three scores can be summed to a total score. High values imply high impairment of the orthopaedic status.

Subjective physical functioning was assessed by the German version of the *Western Ontario und McMaster Universities (WOMAC) Osteoarthritis Index* and the *Haemophilia Activities List (HAL)*. The *WOMAC* is a questionnaire to assess symptoms and physical limitations in patients with knee and/or hip osteoarthritis [15,16]. It consists of three scales (pain, stiffness, function), which can also be added to a total score. In the current study, questions had to be answered concerning only the knees, as these are one of the most commonly affected joints in haemophilic arthropathy [17]. The *HAL* is a 42-item questionnaire, which assesses haemophilia-specific functioning of daily activities in seven dimensions ('lying/sitting/kneeling/standing', 'functions of the legs', 'functions of the arms', 'use of transportation', 'self-care', 'household tasks' and 'leisure activities and sport') and a total score [13]. Component scores can be calculated for upper extremity, basic lower extremity and complex lower extremity activities as well as for total activities. High values imply poor functional health status.

Health-Related Quality of Life (HRQoL) was assessed with two questionnaires: *SF-36 Health Survey (SF-36) and Haem-A-QoL*. The *SF-36* is a generic HRQoL instrument consisting of 36 items pertaining to eight dimensions ('physical functioning', 'role physical functioning', 'bodily pain', 'general health perception', 'vitality', 'social functioning', 'role emotional functioning' and 'mental health') [18,19], which can be summarized to a physical component score (PCS) and a mental component score (MCS) [20]. High values imply a good QoL. The *Haem-A-QoL* is a disease-specific HRQoL instrument for adult haemophilic patients, with 46 items pertaining to ten dimensions ('physical health', 'feelings', 'view', 'sport/leisure', 'work/school', 'dealing', 'treatment', 'future', 'family planning' and 'sexuality') and one total score with high values implying high impairments in patients' HRQoL [21,22].

Data analysis

All statistical analyses were performed using the SPSS program version 15. For psychometric testing of the HEP-Test-Q, information retained from patients' evaluation and results gathered from the preliminary psychometric testing were analysed to decide if an item had to be rejected, modified or retained using the following five criteria:

1. *Missing values* (items which were not answered by more than 10% of respondents should be deleted),
2. *Item difficulty* (items which are not discriminating between respondents should be deleted; e.g. items endorsed only by 20% of respondents or items agreed to by more than 80% of respondents),
3. *Item-total-correlation* (items should be omitted if the correlation of the item with the scale is $<r = 0.30$),
4. *Changes in alpha* (Cronbach's alpha is an indicator of the reliability of a scale and should be at least $\alpha = 0.70$. Deleting a poorly performing item may lead to an increase in Cronbach's alpha) and
5. *Evaluation* (judgements in terms of comprehensibility and relevance from the evaluation were used. If more than 10% of respondents could not understand an item and if more than 20% did not find an item relevant, the respective item should be omitted). A more stringent criterion for comprehensibility was chosen compared with relevance as we considered clarity absolutely critical.

After revision of the questionnaire, corresponding items were recoded, where a higher value indicates better physical performance. Descriptive data are shown as frequency distribution in percentage or as mean (M) ± standard deviation (SD) and range. Data were tested for normal distribution.

Further psychometric testing included testing of reliability and validity. Reliability was calculated for internal consistency (Cronbach's alpha) and test-retest reliability. Convergent validity was determined by means of the Pearson correlation coefficients comparing the HEP-Test-Q scales with objective measures, namely the maximal isometric muscle strength and the OJS as well as with subjective measures, namely the WOMAC, the HAL, the SF-36 and the Haem-A-QoL. Different measures have been included for pilot-testing and field testing, which is because of different project phases of the HEP in which the newly developed HEP-Test-Q was psychometrically tested (see Table 1). The maximal isometric muscle strength and the WOMAC have

Table 1. Measures included in pilot and field testing for convergent validity analysis.

Measure	Parameter	Pilot testing	Field testing
Physical functioning			
Objective			
Knee extension by SCHNELL™	Maximal isometric strength in M. quadriceps femoris	x	–
OJS	Joint status	x	x
Subjective			
WOMAC	Degree of knee osteoarthrosis	x	–
HAL	Physical functioning of daily living	–	x
Health-related Quality of Life			
SF-36 Health survey	Generic quality of life	x	x
Haem-A-QoL	Haemophilia-specific quality of life	x	x

OJS, Orthopaedic joint score; WOMAC, Western Ontario and McMaster Universities Osteoarthritis Index; HAL, Haemophilia activities list.

not been included in the field testing of the HEP-Test-Q as in this project phase of the HEP, they were substituted with other objective and subjective measures considered more relevant and appropriate.

For discriminant validity, patients were classified into different clinical subgroups (age, BMI, bleeding episodes, presence of viral infections and activity level). These subgroup analyses were examined by Student's *t*-test. $P \leq 0.05$ was defined as significant.

Results

Pilot testing

Feasibility testing of the preliminary version Twenty-four PWHs evaluated the preliminary version with 32 items of the HEP-Test-Q on a visual analogue scale ranging from 0 'poor' to 100 'excellent' with a mean value of 57.5 ± 13.5 (10–77). They found the questionnaire easy to fill in 22.4 ± 12.7 (5–59) (0: 'very simple' to 100: 'very difficult') and considered the items relevant/important for their disease 66.8 ± 19.6 (12–98) (0: 'not important' to 100: 'very important'). It took about 14.4 ± 9.6 min (3–40) to fill in the questionnaire.

Item level analysis Seven items did not discriminate between persons (only 20% endorsed or >80% agreed). In five items, the item-total correlation was below the critical value of $r = 0.30$ and Cronbach's alpha would have increased by deleting those items (see Table 2).

Evaluation Only three items out of 32 items were considered not important by more then 20% of patients (e.g. 'I was afraid of injuring myself', 'I was self-confident'). None of the items was incomprehensible (see Table 2).

Based on patients' evaluation and item level analysis, nine of 32 items were eliminated and five items were moved to another scale because of higher correlations with other scales to achieve a higher homogeneity of items. On recommendations of patients, two items were added ('My physical activity was affected because of chronic pain', 'I had problems walking downstairs'), which were included in the field test version.

Psychometric testing pilot testing Reliability testing in terms of Cronbach's alpha revealed values ranging from $\alpha = 0.82$ (general questions) to $\alpha = 0.90$ (endurance) and for the total score of $\alpha = 0.95$ for the revised version of the HEP-Test-Q with 23 items. Test-retest correlation ranged from $r = 0.67$ (general questions) to $r = 0.92$ (strength & coordination). Convergent validity was tested with objective measures, namely the maximal isometric muscle strength and the OJS, as well as with subjective measures namely the WOMAC, the SF-36 and the Haem-A-QoL. Convergent validity showed high correlation between the total score of the HEP-Test-Q and the PCS of the SF-36 ($r = 0.75$, $P < 0.001$), the isometric muscle strength ($r = 0.71$, $P < 0.001$), the WOMAC and the OJS ($r = 0.63$, $P < 0.001$) and the Haem-A-QoL ($r = 0.61$, $P < 0.01$). No significant correlation was found with the HEP-Test-Q total score and the MCS of the SF-36. For discriminant validity, significant differences were found for age ($P < 0.015$), hepatitis B ($P < 0.045$) and patients with different orthopaedic conditions ($P < 0.008$).

Table 2. Item omission, reassignment and new items after evaluation and psychometric testing of the preliminary version of the HEP-Test-Q (32 items, $n = 24$).

		Psychometric testing				Evaluation		Handling
Dimension	Item label	Missing value (>10.0%)*	Difficulty index (>20.0%)*	Item-total correlation ($r < 0.30$)*	Alpha if item deleted	Not important (>20.0%)*	Incomprehensible (>10.0%)*	Item omission, reassignment or new item
Physical status	How do you evaluate your actual physical activity?	–	–	–	–	–	–	Assignment to 'strength & coordination'
Mobility	It was easy for me to shave, comb and put on a sweater	–	50.0	0.21	↑	–	–	Omission
	I had problems with putting on trousers or shoes	–	33.3	–	–	–	–	Omission
	My physical activity was affected because of chronic pain**							New item
Strength & coordination	I had problems walking downstairs**							New item
Endurance	I could carry out/perform moderate activities (e.g. take strolls)	–	29.2	–	–	–	–	Omission
	I could carry out/perform very exhausting activities (e.g. jogging, mountain biking)	–	75.0	–	–	29.1	–	Omission
Body perception	I listened to my body	–	20.8	−0.14	↑	–	–	Omission
	I paid attention to my posture	–	–	−0.00	↑	–	–	Omission
	I paid attention to signals from my body	–	12.5	−0.00	↑	–	–	Omission
	I was afraid of injuring/hurting myself	–	20.8	–	–	20.8	–	Omission
	I perceived bad posture	–	–	0.03	↑	–	–	Omission
General questions	My physical activity was affected because of pain	–	–	–	–	–	–	Assignment to 'mobility'
	I did a lot with others	–	–	–	–	–	–	Assignment to 'endurance'
	I was self-confident	–	–	–	–	21.7	–	Assignment to 'body perception'
	I was physically more active than usual	–	–	–	–	–	–	Assignment to 'endurance'

*Recommended minimum requirement; ↑alpha increases for scale, if item omitted; **No data for psychometric testing and evaluation, since item was added after revision.

Field testing

After adding two items (see Table 2), the revised HEP-Test-Q consisted of 25 items pertaining to four dimensions 'mobility', 'strength & coordination', 'endurance' and 'body perception' as well as one single item, which assessed changes in physical activity. The HEP-Test-Q was field tested in 43 haemophilic patients. More detailed information is provided in Table 3 about clinical characteristics of these patients.

Table 3. Clinical data of haemophilic patients in field testing (n = 43).

Clinical characteristic		n	Frequency distribution (%)
Type of haemophilia	A	40	93.0
	B	3	7.0
Severity (FVIII/IX-activity-level)	Severe (<1%)	38	88.4
	Moderate (1–5%)	3	7.0
	Mild (>5%)	2	4.6
Inhibitors	Yes	3	7.0
	No	40	93.0
HAV	Yes	10	23.3
	No	31	72.1
	Data not available	2	4.7
HBV	Yes	17	39.5
	No	23	53.5
	Data not available	3	7.0
HCV	Yes	28	65.1
	No	14	32.6
	Data not available	1	2.3
HIV	Yes	6	14.0
	No	37	86.0
Treatment	Prophylaxis	21	48.8
	On-demand	22	51.2
Activity level	Not active	20	46.5
	≥1 times per week	23	53.5
Number of bleeds in the last 12 months	Mean ± SD (range)	6.4 ± 6.0 (0–24)	
Number of target joints*	Mean ± SD (range)	0.6 ± 0.8 (0–3)	

*Target joint was defined as >3 bleeds in the same joint in the last 12 months.

Psychometric testing field testing

Reliability The internal consistency (Cronbach's alpha) of the revised HEP-Test-Q ranged from $\alpha = 0.85$ for the dimension 'body perception' to $\alpha = 0.92$ for the dimension 'strength & coordination'. The reliability for the total score was $\alpha = 0.96$ (see Table 4). All values were above the critical value of $\alpha = 0.70$.

Of 43 subjects, 34 filled in the revised HEP-Test-Q (79%) again after 18 days in average (range, 6–48). The test-retest reliability showed high correlations between $r = 0.75$ and 0.93 for all dimensions and $r = 0.90$ for the total score ($P \leq 0.001$) (see Table 4).

Validity

Convergent validity Convergent validity testing showed quite high correlations with the disease-specific QoL instrument (Haem-A-QoL), the physical

Table 4. Scale structure, internal consistency (alpha) and test-retest correlation for the revised version of the HEP-Test-Q (25 items, n = 43; n = 34 for test-retest).

Dimension	No. items	M	SD	Min	Max	Alpha	Test-retest correlation
Mobility	4	13.1	3.5	5	20	0.87	0.81***
Strength & coordination	8	24.1	8.0	10	37	0.92	0.93***
Endurance	8	24.0	6.9	11	37	0.87	0.78***
Body perception	5	16.4	4.0	6	24	0.85	0.75***
Total score	25	77.6	20.3	32	118	0.96	0.90***

***$P \leq 0.001$.

HEP-Test-Q Dimensions	OJS	HAL	PCS (SF-36)	MCS (SF-36)	Haem-A-QoL
Mobility	−0.40**	0.65***	0.65***	0.42**	−0.75***
Strength & coordination	−0.48***	0.76***	0.70***	0.56***	−0.75***
Endurance	−0.51***	0.76***	0.69***	0.56***	−0.78***
Body perception	−0.23	0.45**	0.45**	0.70***	−0.64***
Total score	−0.48***	0.76***	0.71***	0.62***	−0.82***

Table 5. Convergent validity for the revised version of the HEP-Test-Q (25 items, $n = 43$) calculated by Pearson correlations (r).

$^{**}P \leq 0.01$; $^{***}P \leq 0.001$.

performance scale of daily life (HAL) and the PCS of the SF-36. No significant correlation was found between 'body perception' and the OJS (see Table 5).

Discriminant validity Type of haemophilia, severity, presence of inhibitors and HIV were distributed too one-sided (see Table 3) and that is the reason why they were not considered for subgroup analysis. Significant differences were demonstrated for age, hepatitis A and hepatitis B and number of target joints (see Table 6). No significant differences were detected for HCV infection, treatment modality, activity level or number of bleed.

Discussion and conclusion

HEP-Test-Q is a short and well-accepted questionnaire, which assesses subjective physical performance of haemophilic subjects. It could be combined with objective assessments to reveal aspects such as 'body perception' (e.g. 'I felt confident in my body'), which cannot easily be measured objectively. HEP-Test-Q was developed in different phases including item collection by experts in the field of sports medicine, questionnaire development, pilot testing with preliminary psychometric testing and evaluation of items by patients, revision of the questionnaire and field testing. Psychometric testing revealed excellent characteristics for reliability and validity, which will be further tested in a bigger sample. High correlations were found between the total scores of the HEP-Test-Q and the HAL, another subjective measure assessing physical functioning in haemophilic patients. However, correlation for the subscale 'body perception' of the HEP-Test-Q and the total HAL score were much lower, indicating that these instruments assess different aspects of physical functioning such as activities of daily living in the HAL in terms of 'self-care', 'household task' and 'use of transportation' and physical performance such as 'mobility', 'strength & coordination' and 'endurance' in the HEP-Test-Q. Correlations among subjective measures were as high as expected; however, we also found high correlations between the HEP-Test-Q and objective measures such as the isometric muscle strength and the OJS. No significant correlation was

Table 6. Discriminant validity for the revised version of the HEP-Test-Q (25 items, $n = 43$) for transformed scales (0–100, with high scores indicating good physical performance).

Subgroup			Mobility		Strength & coordination		Endurance		Body perception		Total score	
Age	≤40	$n = 12$	61.5	*ns*	69.8	0.000	63.3	0.009	66.3	*ns*	65.7	0.001
	>40	$n = 31$	55.2		42.7		44.8		53.2		47.5	
BMI (kg m^{-2})	≤25	$n = 22$	59.1	*ns*	58.2	0.030	55.8	*ns*	61.1	*ns*	58.2	*ns*
	>25	$n = 21$	54.8		42.0		43.8		52.4		46.7	
HAV	No	$n = 31$	57.9	*ns*	55.3	0.036	55.2	0.015	60.2	*ns*	56.7	0.050
	Yes	$n = 10$	55.0		36.3		36.6		50.0		42.1	
HBV	No	$n = 23$	65.8	0.006	61.4	0.001	58.3	0.008	64.1	0.014	61.7	0.002
	Yes	$n = 17$	46.3		36.2		40.3		48.5		41.6	
HCV	No	$n = 14$	63.4	*ns*	59.6	*ns*	53.8	*ns*	60.4	*ns*	58.5	*ns*
	Yes	$n = 28$	54.2		45.6		47.9		54.8		49.6	
Treatment	Pro	$n = 21$	53.9	*ns*	47.0	*ns*	47.0	*ns*	55.5	*ns*	49.8	*ns*
	On	$n = 22$	59.9		53.4		52.7		58.2		55.2	
Activity level per week	0	$n = 20$	51.6	*ns*	47.2	*ns*	47.0	*ns*	56.3	*ns*	49.7	*ns*
	≥1	$n = 23$	61.7		53.0		52.4		57.4		55.1	
No. of bleedings per year	<5	$n = 18$	53.5	*ns*	44.8	*ns*	48.3	*ns*	55.3	*ns*	49.4	*ns*
	≥5	$n = 22$	62.2		56.8		53.4		58.2		56.9	
No. target joints	0	$n = 23$	51.9	*ns*	41.7	0.017	45.0	*ns*	52.6	*ns*	46.6	0.042
	≥1	$n = 12$	67.2		63.0		57.6		61.7		61.7	

Pro, prophylaxis; On, on-demand.

found for the subscale 'body perception' and the OJS, which demonstrates that subjective measures are important to detect aspects, which cannot easily be measured with objective instruments. The HEP-Test-Q was able to discriminate between clinical subgroups revealing better physical functioning in younger patients and patients without infections.

Originally, the instrument was intended to be a project-specific measure assessing aspects of physical functioning trained in the HEP. By contrast, the HEP-Test-Q proved now to be an instrument, which can be used in general for the assessment of subjective physical performance, but still needs cross-cultural validation in larger samples.

The item collection in the development phase of the HEP-Test-Q was based only on interviews with sports scientists and did not incorporate a patient-centric approach, which could be criticized. On the other hand, during the evaluation phase, patients were asked if any aspects were missing and to make suggestions for additional items. Thanks to this approach, two additional items could be included in the final HEP-Test-Q.

Critical was the low number of participants, which is because haemophilia is a rare disease with a low prevalence. Consequently, there is a limited number of participants in the HEP. The HEP-Test-Q is a short and practical instrument, which can be used in routine clinical practice or can be implemented in clinical trials.

Mainly objective measures are available for the assessment of physical functioning in haemophilic patients such as the OJS for adult haemophiliacs [14], the paediatric Haemophilia Joint Health Score [23] or the Functional Independence Score [24]. Some subjective measures were used for the assessment of physical functioning in haemophilic patients such as the arthritis-specific WOMAC [25] and AIMS-2 [26,27], but these measures were not specific for their disease.

The HAL, the first haemophilia-specific questionnaire for the subjective assessment of physical functioning, was only recently developed [13]. Among other things, HAL focuses on aspects related to activities of daily living such as use of transportation, self-care and household tasks. In contrast, the HEP-Test-Q has a different structure; it is based on motoric abilities such as mobility, strength, coordination and endurance. Based on the assessment of deficits in motoric abilities, attention specifically to sports therapeutic contents could be applied. As the risk of injury of haemophilic patients during speed training velocity (one of the motoric abilities) was not considered in the HEP and consequently not in the HEP-Test-Q, which would exceed the benefits of sports therapeutic programmes. A great focus was directed towards the component 'body perception', which is essential for haemophilic patients. In our opinion, training of body perception at the beginning of a sports therapy can help increase the awareness in haemophilic patients of their body postures, their movements and consequently, their body scheme.

The HAL was included in the current study for convergent validity testing of the HEP-Test-Q showing high correlation for most of the dimensions, except for the HEP-Test-Q subscale 'body perception', which demonstrates that both instruments assess similar, but not identical aspects. Body perception is an important aspect for haemophilic patients for their interoception[1] to recognize flawed body posture, which needs to be corrected and to realize impairments in their physical functioning [8,29]. The HEP-Test-Q is the only instrument attempting to measure subjective body perception in haemophilic patients.

A comparison between HEP participants and healthy age-matched controls as well as a comparison with haemophilic patients not attending the HEP was performed, which will be described in a future paper. A version for children is currently under development.

The sensitivity of the HEP-Test-Q will be tested in the future within the framework of the 2-year follow-up period of the HEP programme. Training effects will be assessed subjectively with the HEP-Test-Q, together with objective measures regarding mobility, coordination and endurance.

An English version of the HEP-Test-Q was developed, which will be included in an observational study, assessing QoL and physical activities in haemophilic patients in the UK (EIS Study).

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Disclosures

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

[1]'Interoception is synonymous with sensory-perceptual process for events occurring inside the body, including visceral perception (i.e. conscious awareness of visceral function)' [28].

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