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The Paediatric Haemophilia Activities List (pedHAL) in routine assessment: changes over time, child-parent agreement and informative domains

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Abstract

Introduction: The Paediatric Haemophilia Activities List (pedHAL) assesses self-reported limitations in activities and participation in children with haemophilia.

Aim: To assess longitudinal changes, child-parent agreement and to identify which pedHAL domains yielded most information in boys with access to early prophylaxis.

Methods: The pedHAL (53 items, 7 domains, optimum 100) was completed annually at the Van Creveldkliniek by boys aged 4-18 years with moderate/severe haemophilia and their parents. Development of the pedHAL in relation to bleeds, changes per domain over 3-5 years, child-parent agreement (% difference child-parent≤|5|) per domain and domain scores (limitations defined as ≤ 95) were determined.

Results: Seventy-three patients and their parents (92% severe haemophilia, median age 13.1 years [range 5.4;18.0]) completed ≥1 pedHAL. Median (IQR) pedHAL sum score was 99.5 (95.2;100.0) for children and 99.6 (95.8;100.0) for parents. If patients scored >95 and had no joint and/or muscle bleed, 90.9% of the patients scored >95 at the next assessment. The median change in sum score was 0.0 for both the 3- and 5-year interval. Child-parent agreement varied between domains from 92% ('self-care') to 71% ('sitting/kneeling/standing'). Most limitations were reported in the domains 'sitting/kneeling/standing', 'functions of the legs' and 'leisure activities and sports.'

Conclusion: In routine clinical practice in Dutch children on prophylaxis, pedHAL scores were high and remained stable in 3-5 years at group level. In individual patients without joint and/or muscle bleeds, pedHAL scores remained high after 1 year. Child-parent agreement was not optimal which indicated that both child report and parent proxy should be reported.

KEYWORDS

activities, child-parent agreement, participation, patient-reported outcome, questionnaire

1 | INTRODUCTION

The Paediatric Haemophilia Activities List (pedHAL) assesses self-reported limitations that children and youth with haemophilia (4-18 years) experience in various activities.[1] The pedHAL measures activities and participation according to the International Classification of Functioning, Disability and Health for Children and Youth (ICF-CY).[2] It was directly derived from the Haemophilia Activities List for adults [3,4] and subsequently validated.[1,5] The pedHAL includes 53 items, distributed over seven domains: 'sitting/kneeling/standing', 'functions of the legs', 'functions of the arms', 'use of transportation', 'self-care', 'household tasks' and 'leisure activities and sports'. The pedHAL is recommended for both research purposes and clinical management of individual patients.[6]

Regular outcome assessment should be part of regular care. However, the optimal frequency of measuring activities and participation is unknown. In addition, some clinicians and researchers reported on items which seemed redundant and some patients experienced the pedHAL as a long questionnaire. Vigorous testing should follow development of a new outcome measure and is not yet performed for the pedHAL. A pilot test in a small sample (n = 32) was performed to describe the score distributions, child-parent agreement, test-retest reproducibility and construct validity of the pedHAL.[1] This first report showed disagreement between patients and their parents in the domain of household tasks and ceiling effects in all pedHAL domains. Field testing plays a role in defining a definitive version of the questionnaire with the minimum number of meaningful items.[7] As a first step, meaningful domains can be explored.

In this field test in Dutch patients with access to early prophylaxis, we explored the optimal frequency of administering the pedHAL and assessed child-parent agreement. In addition, we identified which pedHAL domains yielded most information in Dutch patients.

Key Points

- In routine clinical practice in Dutch children on prophylaxis, pedHAL scores were high and remained stable in 3-5 years.
- Annual pedHAL assessment has limited clinical value in patients without limitations in activities and participation and without joint and/or muscle bleeds.
- Both child report and parent proxy should be reported for the pedHAL.

2 | MATERIALS AND METHODS

2.1 | Study design and study population

This study was a single-centre observational field test of the pedHAL. Since 2010, completion of this list is part of the routine assessment of severe/moderate haemophilia patients aged 4-18 treated at the Van Creveldkliniek in Utrecht, the Netherlands. Children and their parents who completed ≥1 pedHAL during routine assessment were included. For children aged 4-7 years, only parents completed the pedHAL. All pedHAL data collected between October 2010 and June 2017 were included, except for those forms in which more than half of the items was missing.

All the data, including patient characteristics and joint and muscle bleeds, were obtained from the electronic medical files. The Medical Research Ethical Committee (MREC) of the University Medical Center Utrecht reviewed the study (protocol number 17-591/C).

2.2 | Measurements

The pedHAL assesses self-reported limitations in activities and participation in children and youth with haemophilia and their parents. It consists of 53 items, distributed over seven domains, including a patient version (8-18 years) and parent version (4-18 years). The pedHAL was administered as a paper questionnaire.

Patients score the items on a 6-point Likert scale ('impossible', 'always', 'usually', 'sometimes', 'almost never' and 'never'), with a 'not applicable (N/A)' scoring option. Domain scores and sum scores are converted to a normalized domain score ranging from 0 (worst possible functional abilities) to 100 (best possible functional abilities). If more than half of the items of a domain were missing or scored 'N/A', no valid domain score was calculated. If more than half of the items were missing or scored 'N/A', no valid total score was calculated.

Patient characteristics analysed included age at pedHAL assessment, type of haemophilia (A or B), severity of the disease (moderate [factor VIII/IX activity 0.01-0.05 IU/mL] or severe [factor VIII/IX activity <0.01 IU/mL]) and clotting factor regimens (prophylaxis yes/no and age start prophylaxis), as well as the results of routine annual Haemophilia Joint Health Score ($HJHS_{2.1}$) for elbows, knees and ankles (optimum total score 0, worst 124 points).[8] Joint and muscle bleeds documented by clinicians were counted during the periods that pedHAL data were available.

2.3 | Statistical analyses

Patient characteristics were presented as proportions or medians (interquartile ranges [IQR:P25;P75]). Descriptive analyses (median, IQR, range, mean and standard deviation [SD]) were performed for the sum score for the most recently completed pedHAL by patients and parents. Differences in age and HJHS total score for children with pedHAL scores ≤95 points vs pedHAL scores >95 points were tested with a Mann-Whitney *U* test.

Change in pedHAL between the first and second pedHAL sum score (time between pedHAL: 6-18 months) was shown for patients without limitations in activities and participation (>95) and with/without joint and/or muscle bleeds. In addition, proportions of scores which deteriorated by more than 5 points were evaluated after three (2.5-3.5) and 5 (4.5-5.5) years of follow-up. The number of joint and/

or muscle bleeds was compared according to deterioration of the pedHAL with descriptive analyses and Mann-Whitney *U* tests. On group level, change scores of the pedHAL sum and domain scores (*follow-up–baseline*) were evaluated after 3 (2.5-3.5) and 5 (4.5-5.5) years of follow-up. Wilcoxon signed-rank tests were used to test differences in scores between baseline and follow-up. Because most data were available from questionnaires completed by the parents, follow-up analyses were performed on the parent data.

Patient-parent agreement was evaluated per domain and for the sum score with descriptive statistics due to non-normally distributed data. Scores were divided in three categories (>95, 90-95 and < 90 points), and for every category, the numbers and proportions of patients and parents were presented to show agreement in scores. In addition, the proportion of the patients and parents with similar scores (difference child-parent: ≤|5|) was assessed. Wilcoxon signed-rank tests were used to test differences in scores between patients and parents.[9]

Descriptive analyses (median, IQR, range, mean and standard deviation [SD]) were performed per domain of the pedHAL for the most recently completed pedHAL by patients and parents. Based on reported limits of agreement (LoA) of test-retest data,[1] limitations in activities and participation were defined as ≤95 points for domain and sum scores.

3 | RESULTS

3.1 | Patient characteristics and pedHAL sum scores

Seventy-three children with haemophilia A or B were included in this study. All eligible patients have completed at least one pedHAL, except from one patient with severe haemophilia and comorbidities that limited his capacity to complete the pedHAL. Patient characteristics are shown in Table 1. Their median age at the time of completing the last pedHAL was 13.1 years (range 5.4;18.0). Most patients had severe haemophilia (91.8%). Median annual bleeding rate was 1.3 (range 0.0;4.8). The median age (IQR) of start with prophylaxis of clotting factor was 1.9 (1.3;2.8) years. One of 67 severe haemophilia patients had a positive inhibitor titre and received immune tolerance induction (ITI) without prophylaxis with bypassing agents, and 19 were ex-inhibitor patients. None of the six moderate haemophilia patients had a history of inhibitors. Joint health was excellent with a median HJHS score of 0.0 points (range 0.0;11.0) at the time of completing the last pedHAL.

TABLE 1 Patient characteristics

Patient characteristics (n = 73)	Median (IQR) or n (%)
Age (y)	13.1 (10.3; 15.6)
Haemophilia A	64 (88)
Severe haemophilia	67 (92)
Annual bleeding rate[a]	1.3 (0.8; 2.5)
On prophylactic replacement therapy	68 (93)
Inhibitor	
Current	1 (1)
Former	19 (26)
Never	53 (73)
HJHS total score (version 2.1)	0.0 (0.0; 2.0)

Abbreviation: HJHS = Haemophilia Joint Health Score.
[a]n = 68 (with ≥ 1 pedHAL assessment).

At the time of the last completed pedHAL, children had a median (IQR) sum score of 99.5 (95.2;100.0), and their parents had a median (IQR) sum score of 99.6 (95.8;100.0) (Table 2). 'Positive' pedHAL sum scores (≤95 points) were observed in a quarter of the children (23.8%) and parents (23.3%). Age was similar between patients with pedHAL scores ≤95 points and >95 points. HJHS scores were higher in patients with pedHAL scores ≤95 points (median 2.0 [IQR: 0.0;4.0] vs median 0.0 [IQR: 0.0;1.0], P = .044). All patients with moderate haemophilia had a pedHAL sum score >95 points.

3.2 | Changes of the PedHAL over time

3.2.1 | Short-term changes according to reported bleeds

About 90.9% of patients with a pedHAL sum score >95 at first assessment and no joint and/or muscle bleeds during follow-up maintained a pedHAL sum score >95 over median (IQR) 1.0 (0.9-1.2) years (see Figure 1). The pedHAL sum score after 6-18 months for the patient who had a pedHAL ≤95 in the absence of joint and/or muscle bleeds (n = 1) was 93.7; the median (IQR) pedHAL sum score after 6-18 months for the patients who had a pedHAL ≤95 and reported joint and/or muscle bleeds (n = 6) was 85.5 (74.3;93.0).

3.2.2 | Changes over 3 years

Changes over 3 years were assessed in 49 parents. Median (IQR) age of the children was 9.7 (7.0;12.1) at the first assessment. After 3 years, pedHAL scores deteriorated in 18.4%. There was a trend of more joint and/or muscle bleeds in children with a deteriorated pedHAL score than in children without a deteriorated pedHAL score (median number of joint and/or muscle bleeds [IQR] 7.0 [3.0-12.5] vs 3.0 [2.0-6.0], P = .065). The median (IQR) sum scores were similar at baseline (100.0 [96.4;100.0]) and three years later (99.6 [94.9;100.0]): median (IQR) change sum score after three year follow-up was 0.0 (−0.9;+0.9).

3.2.3 | Changes over 5 years

Changes over 5 years were assessed in 35 parents. Median (IQR) age of the children was 9.3 (6.9;10.6) at the first assessment. After 5 years, pedHAL scores deteriorated in 14.3%. The number of joint and/or muscle bleeds was higher in children with a deteriorated pedHAL score than in children without a deteriorated pedHAL score (median number of joint and/or muscle bleeds [IQR] 5.0 [4.0-12.3] vs

TABLE 2 Score distribution pedHAL per domain

	Children (n = 63)				Parents (n = 73)			
Domain	Median (IQR)	Min	Max	Score ≤95 (%)	Median (IQR)	Min	Max	Score ≤95 (%)
Sitting/kneeling/standing	100 (94.0;100)	66.0	100	29	100 (96.0;100)	56.0	100	22
Functions of the legs	100 (94.5;100)	62.0	100	29	100 (96.4;100)	14.6	100	23
Functions of the arms	100 (96.0;100)	68.0	100	19	100 (100;100)	60.0	100	15
Use of transport	100 (100.0;100)	60.0	100	10	100 (100;100)	60.0	100	16
Self-care	100 (100.0;100)	75.6	100	13	100 (100;100)	64.4	100	14
Household tasks	100 (100.0;100)	60.0	100	14	100 (100;100)	60.0	100	13
Leisure activities and sports	100 (96.4;100)	54.3	100	20	100 (95.1;100)	7.3	100	25
Sum score	99.5 (95.2;100)	76.3	100	24	99.6 (95.8;100)	49.6	100	23

Note: Descriptive analyses were performed according complete case analysis.

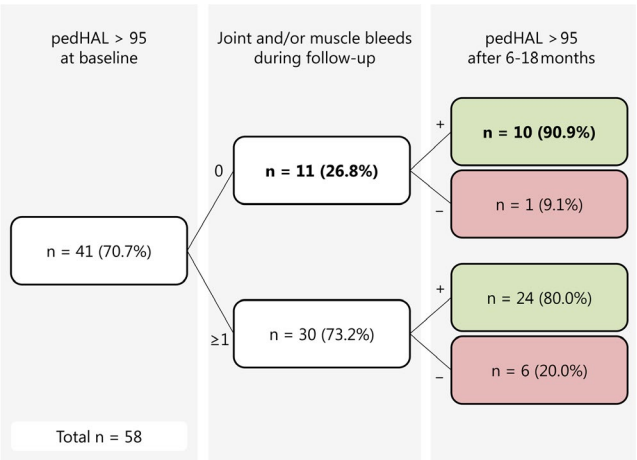

FIGURE 1 Changes of pedHAL score within one year (6-18 mo) of follow-up according to bleeding. + present; − absent [Colour figure can be viewed at wileyonlinelibrary.com]

15.0 [8.0-24.5], $P = .024$). The median (IQR) sum scores were similar at baseline (100.0 [97.3;100.0]) and 5 years later (100.0 [98.8;100.0]): median (IQR) change sum score after 5 year follow-up was 0.0 (−1.0;+0.9).

For all seven domains, median change scores were also 0.0 after 3 and 5 years. Repeat analyses of the pedHAL scores completed by children yielded similar results.

3.3 | Child-parent agreement

Table 3 shows descriptive analyses of agreement of the domain scores and sum score of the pedHAL in 63 children and parents. At domain level, child-parent agreement (difference child-parent ≤|5|) varied across domains: agreement was highest in the domain 'self-care' (92%) and lowest in the domain 'sitting/kneeling/standing' (71%). In addition, at domain level children and parents scored >95 points in 79% ('sitting/kneeling/standing') to 98% ('self-care'). For the sum score, child-parent agreement (difference child-parent ≤|5|) was 81%. If children scored >95 points on the pedHAL, parents scored >95 points in 96%.

3.4 | Score distribution of the pedHAL domains

Table 2 shows the median (IQR) and the range of the domain scores and proportions of sum and domain scores ≤95 points of the last completed pedHALs. Mean scores (SD) are shown in Appendix S1. All domain scores were median 100.0. The domain 'functions of the legs' showed most 'positive' scores (28.6% of children and 23.3% of parents). In children, the domain of 'use of transport' showed least 'positive' scores: in 9.8%. In parents, the domain of 'household tasks' showed least 'positive' scores: in 12.9%.

4 | DISCUSSION

This study showed that Dutch children on early prophylaxis and their parents reported almost no limitations in activities and participation. On group level, after 3 or 5 years follow-up the sum scores and domain scores remained stable. On patient level, in patients without limitations in activities and participation (pedHAL sum score >95) and without joint and/or muscle bleeds, pedHAL scores remained high till the next assessment after median (IQR) 1.0 (0.9-1.2) years. If patients did report limitations, they reported most limitations in the domains 'sitting/kneeling/standing', 'functions of the legs' and 'leisure activities and sports'. Almost no limitations were reported on the pedHAL items in the domains 'functions of the arms', 'use of transportation', 'self-care' and 'household activities'. In addition, child-parent agreement (difference child-parent ≤5) varied across domains from 71% agreement for 'functions of the legs' up to 92% agreement for 'self-care'. The differences indicated that both child report and parent proxy should be reported.

4.1 | Internal and external validity

So far, studies on the psychometric properties of the pedHAL have been limited.[10] The results of the present report were dependent on the choice of cut-off points. The three categories (>95, 90-95 and <90 points), cut-off score of ≤95 (limitations in activities and participation) and cut-off score of more than 5 points for changes in

TABLE 3 Child-parent agreement (n = 63 pairs)

Children	Parents	Sitting/kneeling/standing	Functions of the legs	Functions of the arms	Use of transport[a]	Self-care	Household tasks	Leisure activities and sports	Sum score
>95 n (%)	>95	**38 (79)**	**41 (87)**	**48 (92)**	50 (96)	**55 (98)**	**44 (92)**	**33 (83)**	**46 (96)**
	90 ≤95	6 (13)	2 (4)	1 (2)		1 (2)	1 (2)	1 (3)	2 (4)
	<90	4 (8)	4 (9)	3 (6)	1 (2)		1 (2)	1 (3)	
	N/A				1 (2)		2 (4)	5 (13)	
90 ≤ 95 n ((%)	>95	2 (40)	1 (25)	1 (50)	2 (100)		1 (100)	1 (100)	
	90 ≤95		**2 (50)**	**1 (50)**					**1 (33)**
	<90	3 (60)	1 (25)						2 (67)
	N/A								
<90 n (%)	>95	3 (30)	2 (17)	1 (11)	3 (33)		1 (14)	3 (25)	1 (8)
	90 ≤95		1 (8)	1 (11)		2 (29)	2 (29)	1 (8)	1 (8)
	90	**7 (70)**	**9 (75)**	**6 (67)**	**5 (56)**	**5 (71)**	**4 (57)**	**5 (42)**	**10 (83)**
	N/A			1 (11)	1 (11)			3 (25)	
N/A n (%)	>95						3 (43)	2 (20)	
	90 ≤95							1 (10)	
	<90							2 (20)	
	N/A						**4 (57)**	**5 (50)**	
Pairs with similar scores, difference child-parent ≤5 (%)		**71**	**73**	**84**	**84**	**92**	**81**	**78**	**81**

Note: Number and proportion n (%) of children and parents who scored >95 points, 90 ≤ 95 points, <90 points and N/A for all domains and the sum score. Concordant scores between children and parents are marked in bold.

[a]*P*-value of the Wilcoxon signed-rank order test is significant ($P < .05$) signifying statistically significant discordant scores between children and their parents.

pedHAL were based on reported limits of agreement (LoA) of test-retest data,[1] but are arbitrary. To determine exact changes in limitations measured with the pedHAL, smallest detectable change and minimal important change should be known.

Results show high pedHAL scores, no changes on group level and stable pedHAL scores in patients without limitations and joint and/or muscle bleeds during 1-year follow-up. Although this field test of routine assessments included data of an unselected cohort with a full age range and long follow-up, the results are only applicable to intensively treated patients as patients in the Van Creveldkliniek receive early prophylaxis and intensive treatment.

Children with a deteriorated pedHAL score after three or five years showed more joint and/or muscle bleeds, which is in line with the hypothesis that bleeds results in limitations of activities and participation. This needs to be confirmed in another population.

4.2 | Comparison with other studies

Neither long-term follow-up data of self-reported limitations in activities and participation measured by the pedHAL, nor child-parent agreement has been published until now. This is the first study which showed three to five years of follow-up data and the development of the pedHAL in relation to joint and/or muscle bleeds. Only in a pilot test, child-parent agreement was studied in a small sample of 15 patients with LoA (change in mean score ±95% confidence interval) varying from 0.7 ± 3.4 to 0.7 ± 28.2.[1] Child-parent agreement in other patient-reported outcome measures (PROMs) used in children with haemophilia show different scores on self-reported health-related quality of life between children and their parents for the Canadian Hemophilia Outcome—Kids' Life Assessment Tool (CHO-KLAT). Differences between child and parent proxy scores were also reported in other paediatric PROMs, and the recommendation is to measure both perspectives.[11,12]

The domain and sum scores as reported in the present study in Dutch children were higher than previous reported data in UK, Romania and Lithuania.[5,13,14] Comparable sum scores were reported in a Canadian study.[15] Similar to the present study, the domains 'sitting/kneeling/standing', 'functions of the legs' and 'leisure activities and sports' were the most informative domains,and most ceiling effects were observed in the domains of 'functions of the arms', 'use of transportation', 'self-care' and 'household activities' in English, Romanian and Lithuanian children.[5,13,14] No studies were available about pedHAL scores in children without haemophilia, to compare the limitations to healthy children.

In the adult version of the HAL, the same seven domains are represented and reported HAL data were mostly comparable to data of children. Adults with mild to severe haemophilia from the United States and adults with severe haemophilia from the UK reported most complaints in the domains 'lying down/sitting/kneeling/standing' and 'functions of the legs' and least complaints in the domain 'self-care'.[16,17]

4.3 | Clinical implications and future research

Results suggest that annual pedHAL assessment in clinical care is not necessary in children who have a pedHAL sum score >95 and no joint and/or muscle bleeds during follow-up. In children with lower pedHAL scores and/or bleeds, annual pedHAL assessment is recommended to monitor limitations in activities and participation. At group level, changes were clinically insignificant after 3-5 years. The present data suggest that the frequency of administrating the pedHAL can be lowered to every 3-5 years when studying groups of children with low bleeding rates receiving intensive prophylaxis. However, both child report and parent proxy should be reported because scores differed between children and their parents. These discrepancies are important to discuss with the child and their parents.

Shortening the questionnaire may enhance the feasibility of the pedHAL within the context of multiple outcome assessment in haemophilia care. The present study identified several pedHAL domains which were less informative; especially 'functions of the arms', 'use of transportation', 'self-care' and 'household activities' may be candidates for shortening. However, before deciding on the removal of items, this study should be repeated including data from other paediatric haemophilia populations, including patients with more frequent bleeding and/or extensive arthropathy.

5 | CONCLUSION

This explorative clinical study suggests that annual pedHAL assessment has limited clinical value in patients without limitations in activities and participation (pedHAL sum score >95) and without joint and/or muscle bleeds. Furthermore, both child report and parent proxy should be reported since scores between children and their parents differ (child-parent agreement: 71%-92%). This study revealed little limitations in activities and participation (pedHAL >95:76%) in Dutch children on prophylaxis and pedHAL scores remained stable over three to 5 years at group level. Domain analyses showed that the domains 'sitting/kneeling/standing', 'functions of the legs' and 'leisure activities and sports' are most informative domains in patients receiving early prophylaxis.

DISCLOSURES

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

AUTHOR CONTRIBUTIONS

All authors contributed to the design of the study, IK performed the statistical analyses, IK wrote the first draft of the paper, all authors contributed to interpretation of the data, modification of statistical analyses and the writing of the manuscript.

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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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