The effect of Nordic Walking on joint status, quality of life, physical ability, exercise capacity and pain in adult persons with haemophilia

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Nordic Walking is an exercise form requiring significant energy consumption, but where the use of poles minimizes the risk of injury. The aim of this pilot study was to examine the effect of 3 months of Nordic Walking on males (>40 years of age) with haemophilia, regarding joint function (Haemophilia Joint Health Score), physical ability (Haemophilia Exercise Project - Test-Questionnaire), exercise capacity (6-min walking test), pain (visual analogue scale) and quality of life (the Swedish version of The Short Form Health Survey, SF-36). Pre-interventional and postinterventional scores of above-mentioned parameters were analysed, using Wilcoxon Signed Ranks Test. Eleven participants were recruited to the study. Statistically significant improvements were observed in physical ability (P value: 0.01) and body perception (P value: 0.02). The intervention did not increase number of bleedings or factor consumption. This is the first study ever evaluating Nordic Walking in persons with haemophilia. Our results suggest

Introduction

Persons with haemophilia (PWH) who have not received prophylaxis treatment from childhood, will with age develop haemophilic arthropathy, because of repeated joint bleeds. Abnormal gait and impaired balance is putting these patients at risk of falls, with new bleeding episodes as a consequence [1].

PWH were discouraged to participate in sports until the 1970s, because of the risk of overload and injury. Today, the opinion is changed and it has been reported that isometric, isokinetic and proprioceptive exercise can improve muscle strength [2-5] and bleeding profile [5,6] in PWH. However, the majority of the literature in this field focuses on the benefits of exercise in children and young adults [7,8] rather than the elderly, although preserving an active lifestyle throughout the years is necessary to prevent falls, arthropathy and osteoporosis [9]. Low-intensity aerobic exercise has become highly recommended for older adults with osteoarthritis in the hip or knees [10,11] and may also be suitable for PWH. Nordic Walking activates muscles in the arms, shoulders, abdomen and the back as a result of force applied on walking-poles, resulting in almost twice as much highenergy consumption, compared with regular walking [12]. In addition, balance is improved and pace can be adjusted according to health status.

that Nordic Walking is safe and efficient, also in patients with haemophilic arthropathy. *Blood Coagul Fibrinolysis* 27:467–472 Copyright © 2016 Wolters Kluwer Health, Inc. All rights reserved.

Blood Coagulation and Fibrinolysis 2016, 27:467-472

Keywords: age, arthropathy, exercise, haemophilia, Nordic Walking, physical ability

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Received 16 September 2015 Revised 7 January 2016 Accepted 11 January 2016

The aim of this study was to examine the effect of 3 months of low-intensity training on men (>40 years of age) with moderate to severe haemophilia regarding joint function, physical ability, exercise capacity, pain and quality of life.

Materials and methods Study population

Subjects were recruited from three haemophilia treatment centres in Sweden (Malmoe, Stockholm and Gothenburg). PWH over 40 years of age – who were not able to receive prophylactic treatment in their early childhood and therefore had developed joint problems – diagnosed with either moderate (factor level of 1-5 kIU/l) or severe (factor level <1 kIU/l) haemophilia A or B, were asked for participation during health check-up meetings, by mail or by phone. The possibility of enrolment was also offered at a haemophilia community meeting. Out of the 68 contacted persons, 11 consented to participate. The study was approved by Lund University Ethical Committee.

Study design and procedure

In this interventional study, the PWH were required to perform 30 min of Nordic Walking, at least three times a week, during a period of 3 months and were motivated to continue for an additional 3 months (6 months in total).

Table 1	Included assessments	during the Nordic Walking	study and the time for	each test performance
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Assessment	Baseline	After 3 months	After 6 months
HJHS 2.1 ^ª	Х	х	
SF-36 ^b	х	Х	х
TSK-SV ^c	х	Х	х
HEP-test-Q ^d	х	Х	х
Approximation of current physical activity and daily exercise	х		
Treatment log		Х	
Training log		Х	
6MWT ^{e,f}	х	х	

^a Haemophilia Joint Health Score, version 2.1. ^b Short Form (36) Health Survey. ^cTampa Scale of Kinesiophobia, Swedish version. ^d Haemophilia & Exercise Project – Questionnaire. ^e 6-Min walk test. ^fThe following assessments were performed in connection to the 6MWT: heart rate, blood pressure, body mass index, oxygen saturation, breathlessness, endurance and pain.

The included PWH were equipped with adjustable spring-system walking poles and neck-worn pedometers (Silva). All exercise and use of analgesics was documented in a training-log. The use of factor replacement was documented in a treatment-log. All included PWH were invited to a full-day conference which started with an inspirational lecture, focusing on ways to increase motivation and the importance of physical activity. The participants were trained in the technique of Nordic Walking and the use of pedometers by two physiotherapists. The preintervention questionnaires were also completed in this session (Table 1). Physiotherapists made regular calls every third week throughout the first 3 months of the study to motivate and answer questions regarding the activity. After 3 months, postinterventional values were assessed. The participants were encouraged to continue the Nordic Walking on their own for an additional 3 months. After a total of 6 months, the PWH received the second set of questionnaires by post (Table 1).

Training-log and treatment-log

The PWH documented each workout at home in a personal training-log. The duration, eventual use of analgesics before/after exercise and the number of steps were documented. The exertion was evaluated using the Borg Rating of Perceived Exertion Scale [13]. The pain-level was assessed using the visual analogue scale [14]. The use of factor replacement was documented in a treatment-log. In case of bleeding, the location of the bleed was specified.

In Sweden, patients are routinely asked to document bleedings and factor consumption and send the reports to their respective haemophilia centre, thus, the number of bleeds during the study period could be compared with the annual bleeding rate of the year before the study (2013). To determine whether the intervention caused an increased number of bleedings and hence, a higher treatment cost, the average amount of factor used per bleeding during the study period was calculated. This was then compared with the ABR and factor consumption of 2013 divided by 4 to achieve the estimated bleeding rate for 3 months.

Joint health status

The joint health of the PWH was assessed by Haemophilia Joint Health Score (HJHS) 2.1 [15] before and after the 3 months intervention period. The HJHS 2.1 Total Score ranges from 0 to 124, with higher scores indicating worse joint status.

General exercise capacity

6-min walking test (6MWT) [16] was performed at the start of the study and after three months of the Nordic Walking intervention. Before the 6MWT, measurements of weight, length and blood pressure were taken. Pre- and post-test values of heart rate and oxygen saturation were measured using GE Ohmeda TuffSat pulse oxiometer. Dyspnoea, exertion and pain were assessed using their respective Borg's scales. The modified Borg CR10 Scale from 2010 was used to estimate dyspnoea and pain [17].

Physical ability, quality of life and kinesiophobia

The physical ability, quality of life and kinesiophobia were evaluated before and after the 3 month intervention period as well as at the second follow-up, using the Swedish version of the Haemophilia Exercise Project – Test-Questionnaire (HEP-Test-Q) [18], the Swedish version of The Short Form Health Survey (SF-36) [19] and The Tampa Scale of Kinesiophobia (TSK) [20,21], respectively. In the case of more than 25% missing items within the same HEP-Test-Q domain, the Domain-score and consequently the Total-score were not calculated. A mean value was used in those cases when an item was incorrectly answered by more than one option being chosen.

Statistics

Statistical analysis was performed using the computing environment R [22]. A nonparametric test, Wilcoxon Signed Ranks Test, was used to compare baseline and postinterventional data. A P value of less than 0.05 was considered statistically significant.

Results

Patient characteristics

Eleven males were recruited to the study. One participant was excluded, as he did not hand in the training and

	Type of haemophilia	Treatment type	Location of undergone joint surgery	Physical workout per week (running, gymnastics, ball games, etc.)	Daily exercise per week (walking, cycling and gardening)	Adequate ^a physical activity per week (yes/no
Patient 1	Severe	Prophylaxis	Left ankle	0	1 h	No
Patient 2	Severe	On demand	Both ankles, both knees, both elbows	6 h	7 h	Yes
Patient 3	Moderate	Prophylaxis	None	0	2 h 40 min	Yes
Patient 4	Severe	Prophylaxis	Left ankle, both elbows	0	1 h 10 min	No
Patient 5	Severe	Prophylaxis	None	0	35 h	Yes
Patient 6	Moderate	On demand	Left shoulder	3 h	50 min	Yes
Patient 7	Moderate	On demand	Left ankle, both knees, left hip	0	2 h 40 min	Yes
Patient 8	Severe	Prophylaxis	None	2 h 30 min	0	Yes
Patient 9	Severe	Prophylaxis	None	1 h	2 h 15 min	Yes
Patient 10	Moderate	On demand	Both knees, right hip	0	0	No

Table 2 Baseline demographic characteristics and clinical data

^aAccording to the Swedish national board of health and welfare.

treatment-logs, leaving 10 patients – diagnosed with either moderate (40%) or severe (60%) haemophilia – under study. The median age was 63 years (range: 40–83) and the median body mass index at study inclusion was 24 kg/m^2 (range: 21–37). None of the patients had lung or heart disease. Two participants were treated for hypertension. The patients did not receive prophylactic treatment since childhood; however, all of them have been using factor replacement for the majority of their lifetime. Seven patients had undergone either unilateral or bilateral joint surgery. Four participants regularly performed physical workout at study inclusion (Table 2).

Training intensity and pain during the first 3 months

Four of the 10 participants achieved the goal of performing at least three Nordic Walking workouts per week, during the first 3 months (median: 2.4 workouts/week). The median duration and walking pace of a workout for all 10 males was 43 min (range: 21–93) and 4.0 km/h (range: 1.5–6.5), respectively. The patients estimated the exertion level of a workout to a median of 12, which corresponds to 'fairly light exertion' to 'somewhat hard' (Borg Rating of Perceived Exertion Scale). The painlevel (visual analogue scale) was significantly increased after a workout (P < 0.001). The patients reported a median pain level of 0.7 before exercise (range: 0–8) and 0.8 after exercise (range: 0–7.5). Two patients used analgesics during the intervention period (Table 3).

Factor consumption during the first 3 months

Eight patients regularly used factor replacement as prophylaxis treatment. Five patients used factor replacement during the intervention to specifically treat bleedings (Table 4). Three of these also had a high ABR the year before the study. The factor consumption during the study period was lower than that of a corresponding 3-month period in 2013 (Table 4).

Joint health status and general exercise capacity after 3 months

The HJHS-total score stayed virtually unchanged from baseline (median: 25, range: 2–59) to post-intervention (median: 32, range: 1–50). The average maximal walking distance during 6 min did not change significantly

	Average workouts per week	Average distance per workout (km)	Average workout duration (min)	Average walking pace (km/h)	Average exertion level per workout	Average pain level before workout	Average pain level after workout	Use of analgesics
Patient 1	1.8	2.5 ^a	33	4.6 ^a	13	0.9	0.8	
Patient 2	3.0	3.7	51	4.4	18	0.1	0.1	
Patient 3	2.3	2.6	42	3.8	12	0.9	1.1	
Patient 4	3.0	3.5 ^a	44	4.8 ^a	10	0.1	0.1	
Patient 5	1.0	3.1	50	3.9	12	2.1	3.7	
Patient 6	2.3	4.5 ^a	93	2.9 ^a	11	0.9	1.6	Three times before and once after a workout
Patient 7	1.6	1.0	41	1.5	15	7.3	6.4	
Patient 8	3.0	2.4	36	4.0	12	0.1	0.6	
Patient 9	2.4	6.0	56	6.5	14	3.0	3.9	Three times after a workout
Patient 10	3.1	1.4	21	3.9	13	0.6	0.7	
Median	2.4	3.4	43	4.0	12	0.9	1.0	
Range	1.0-3.1	1.0-6.0	21-93	1.5-6.5	10-18	0.1-7.3	0.1-6.4	

Table 3 Patient-specific summary of training logs. Mean values are given at each patient-specific row. Median values are given at group level

^a The length of steps for these patients has not been registered. Conversion from steps to meters has been done by multiplying the number of steps by 0.71, which is the median length of steps for the other patients.

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	Factor concentrate	Frequency of prophylaxis treatment (days/week)	Units per infusion	No. treated bleedings	Bleeding location	Annual bleeding rate 2013	Units of factor replacement used to treat bleedings during the study period (3 months)	Units of factor treatment used during a 3-month non-study period (according to ABR 2013)
Patient 1	Advate	2.6	2 000	0		NDA	I	1
Patient 2	Advate	0.1	2 000	ю	Back muscles	0	6 000	0
Patient 3	Kogenate	1.8	2 000	0		NDA	I	I
Patient 4	Benefix	3.4	4 000	e	Left elbow	0	12 000	0
Patient 5	Kogenate	2.2	2 000	18	Back muscles, ankle, wrist	120	36 000	60 000
Patient 6	,	None		0		NDA	I	I
Patient 7		None		0		NDA	I	I
Patient 8	Nanofix	0.5	2 000	0		0	I	I
Patient 9	Kogenate	1.8	2 000	-	Left ankle	10	2 000	6 000
Patient 10	Imunine	0.5	2 400 (1 200 at	5	Left knee (four occasions),	11	12 000	7 200
			one occasion)		left ankle (once)			
							TOTAL: 68 000	TOTAL: 73 200

(*P* value: 0.06) from baseline (median: 450 m, range: 315-670 m) to the first follow-up (median: 458 m, range: 363-679 m). Neither did body mass index (*P* value: 0.68), nor blood pressure (systolic *P* value: 0.92, diastolic *P* value: 0.84). The resting oxygen saturation and pain level (before 6 min of walking) at baseline did not change significantly compared with resting levels at the first follow-up (*P* values: 0.06 and 0.24, respectively).

Quality of life, kinesiophobia and physical ability after 3 months

Neither quality of life (*P* value Physical Composite Summary: 0.28, *P* value Mental Composite Summary: 0.56) nor the degree of kinesiophobia (*P* value: 0.12) changed significantly during the 3-month intervention. However, significant changes were observed in physical ability (HEP-Test-Q) (Fig. 1), explicitly in the total-score (*P* value: 0.01) and within the body perception parameter (*P* value: 0.02), which both increased from baseline.

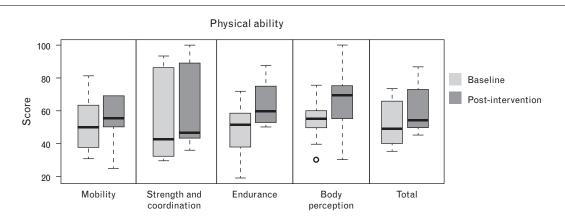
Quality of life, kinesiophobia and physical ability after 6 months

Six participants reported they did not continue to exercise (patients 2-6 and 10), patient 1 did continue with Nordic Walking, patient 8 replaced Nordic Waking with table tennis and two participants were lost to follow-up (patients 7 and 9).

Only six of the participants handed in the second followup questionnaires. The quality of life did not change significantly, when comparing the scores after 6 months to baseline scores (P value Physical Composite Summary: 0.84, P value Mental Composite Summary: 0.56) or to the first follow-up (P value Physical Composite Summary: 0.56, P value Mental Composite Summary: 0.16). The degree of kinesiophobia decreased between baseline and the first follow-up, but increased again at the second follow-up. The changes were not significant (P value for baseline to second follow-up comparison: 0.56, P value for first to second follow-up comparison: 0.14). The total physical ability did not change significantly when comparing baseline levels to the second follow-up (P value: 0.29) or when comparing the first follow-up to the second follow-up (P value: 0.06). However, a significant decrease was observed in the strength and coordination domain (P value: 0.04), when comparing the scores from the first follow-up to the scores from the second follow-up.

Discussion

This pilot study evaluated the effect of 3 months of Nordic Walking on 10 adult PWH. Being the first study of its kind, it indicates that Nordic Walking is a safe exercise form, resulting in improvements in physical ability in general and body perception in particular, despite presence of arthropathy. The intervention did not increase the use of factor replacement or bleeding frequency, compared with pre-interventional treatment



Physical ability scored by HEP-TEST-Q test was documented in 10 PWH at baseline (light grey) and after 3 months of Nordic walking (grey). Patient number 6 did not reach the threshold of 75% non-missing items within the mobility domain; consequently his Mobility-score and Total-score could not be calculated. Median values and ranges are indicated. Significant differences were seen in the total score (P=0.01) as well as in the Body perception domain (P=0.02).

logs. Three of the five participants who experienced bleedings during the study period also had a high ABR the year before the study, indicating it was not primarily the increased exercise causing bleedings.

Regarding exercise capacity, a positive trend (P value: 0.06) was observed in the 6-min walking distance; patients walked on average 8 m longer post-intervention, resulting in 458 m, which is approximately 72% of the reference value in healthy males between 20 and 50 years of age [23]. In addition to the before mentioned significant improvements in physical ability, positive trends with P values less than 0.06 were observed in the strength and coordination and endurance domains. These findings are in line with results from the Haemophilia and Exercise Project, in which adult PWH underwent a 1-year sports therapy program, which resulted in improved performance in the total score and in all domain-scores (except of mobility), compared with inactive and control PWH [24].

Though this pilot study presents promising results, the study has important limitations: the sample size is low, it is not randomized and there were no controls. We believe that the reason we could not show improvements regarding quality of life, joint health status or pain is primarily because of the relatively short intervention period (3 months).

Nordic Walking has previously been shown to improve the health status in a number of patient groups [25]. Several patients in this study expressed that they felt supported by the walking poles, which implies the safety of this exercise form. The patients reported a statistically significant increase in pain after performing exercise; however, the median visual analogue scale pain-level was below 1 (very week pain) and thus, the increase cannot be said to be clinically relevant. Although some of the patients did express that they felt stronger after the first three months of Nordic Walking, only one patient continued the practice. The inactivity between the third and the sixth months of the study seems to have reverted the positive trend in physical ability.

The need of finding a suitable exercise form for PWH is especially apparent in developing countries with insufficient amount of factor replacement, where physiotherapy is necessary as a complement to improve muscle strength and to reduce pain [26]. The encouraging results presented in this study has contributed to the planning of a future study, spanning a longer time period and including more patients, where additional measures to enhance motivation will also be taken. We also want to encourage other centres to organize similar training groups and we would welcome any collaboration on the subject.

Acknowledgements

M.S. analysed the data and wrote the manuscript. E.B. performed the research and contributed in writing of the methods section, Y.S.-A. performed the research, E.B. designed the study and contributed to the writing of the paper, and E.Z. designed the study, performed the research and wrote the paper.

The skills of research nurse Eva Linden, Skane University Hospital, is gratefully acknowledged as well as the help from Valdimir Radulovic, Sahlgrenska University Hospital, Gothenburg and Maria Bruzelius, Karolinska University Hospital with the help to recruit participants to the study.

The authors stated that they had no interests that might be perceived as posing a conflict or bias. The work was supported by an unrestricted grant from Baxter and by an unrestricted grant from Novo Nordisk.

Conflicts of interest

There are no conflicts of interest.

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