

Relationship between fear of movement and physical activity levels in adult hemophilic individuals

Sercan Aykar¹, Filiz Can², Fahri Sahin³

¹Adnan Menderes University, Faculty of Health Sciences, Department of Physiotherapy and Rehabilitation, Aydin, Turkey

²Hacettepe University, Faculty of Physical Therapy and Rehabilitation, Department of Physiotherapy and Rehabilitation, Ankara, Turkey

³Ege Adult Haemophilia and Thrombosis Center, Izmir, Turkey

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Abstract

Aim: Regular physical activity is important to prevent damage and protect joint health in hemophilic individuals. However, hemophilia patients tend to limit their physical activity due to frequent bleeding. There may be some factors, such as fear of movement or kinesiophobia under these attitudes. For this reason, this study aimed to research the relationship between kinesiophobia and physical activity levels in adult hemophilic individuals.

Material and Methods: This study was performed with a retrospective study design. A total of 44 patients included in this study; 33 were hemophilia A, 11 were hemophilia B. All hemophilic individuals were male and between the ages of 18-60 (mean=35.30±12.04). Tampa Scale for Kinesiophobia (TSK) was used to determine fear of movement. To determine the physical activity levels of individuals, the "International Physical Activity Questionnaire-Short Form" was used. The results were analyzed using Spearman correlation analysis and Mann-Whitney U test.

Results: There was no statistical correlation between kinesiophobia degree and physical activity level ($p=0.616$, $r=-0.081$). There was also no significant difference in kinesiophobia scores between hemophilia A and hemophilia B ($z=-0.707$, $p=0.479$). High kinesiophobia levels (TSK score of ≥ 37) were present in 90.9% of patients. According to physical activity levels, 36.4 % of patients had low, 34.1 % had moderate and 29.5 % had high physical activity levels. Hemophilia A patients had higher physical activity levels than hemophilia B ($z=-2.400$, $p=0.016$).

Conclusion: These results showed no correlation between physical activity level and kinesiophobia, but high kinesiophobia rates in adult hemophilic individuals. Hemophilia patients should be educated and encouraged about safe and fearless physical activity.

Keywords: Movement; fear; Hemophilia A; Hemophilia B

INTRODUCTION

Hemophilia is a congenital clotting disorder inherited in an X-linked recessive genetic pattern which leads to a lack or failure of a blood-clotting protein (1). There are two common types of hemophilia: hemophilia A (factor VIII (FVIII) deficiency) and hemophilia B (factor IX (FIX) deficiency). Patients with hemophilia experience various degrees of bleeding due to incomplete coagulation factors that especially affect musculoskeletal structures. Repeated musculoskeletal bleeding may cause hemophilic arthropathy, pain, activity limitation, decreased range of motion and quality of life (2). However, hemophilia B is thought to be associated with lower bleed frequencies and less severe disability (3). There is a variable bleeding

phenotype in the clinical manifestations of hemophilia (4).

The severity of hemophilia and age should not be seen as obstacles to being physically active (5). Moreover, for patients with hemophilia, it has been shown that is worthwhile to start physical activity at an early age. Physical symptoms of inactive behavior (muscle weakness, instability, and limited movement) have been detected in some children with hemophilia and may become worse and more problematic with increasing age (4,6). The World Federation of Hemophilia (WFH) recommends physical activity to enhance the quality of life with improved prophylaxis and hemostasis treatment opportunities (7).

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Corresponding Author: Sercan Aykar, Adnan Menderes University, Faculty of Health Sciences, Department of Physiotherapy and Rehabilitation, Aydin, Turkey **E-mail:** sonal@adu.edu.tr

One of the reasons for physical activity restrictions is "kinesiophobia". Kinesiophobia or fear of movement is defined as an extreme, unreasonable and weaken fear of physical activity and movement (8). The basic concept is the fear of feeling pain or specifically the fear of (re)injury caused by physical activity (9). Tampa Kinesiophobia Scale (TKS) is a commonly used assessment of kinesiophobia in clinical practice (9). It is reported that higher degrees of kinesiophobia are related to higher levels of pain, disability and lower quality of life (10).

There is no conclusive evidence about physical activity levels of hemophilia patients in the literature, but most reports define equal or higher physical activity levels in comparison with healthy controls (11-13). In contrast, a report found decreased levels of physical activity in hemophilic individuals, and that the main indicators of decreased physical activity levels were increased severity of hemophilia and age (14).

Investigating the relation of kinesiophobia and physical activity level would be useful to understand the behavioral responses of the hemophilic individuals, and the differences between hemophilia type. For this reason, the present study aimed to investigate possible relationships between kinesiophobia and level of physical activity in patients with hemophilia. This study also explored the differences between kinesiophobia and physical activity levels of patients with hemophilia A and hemophilia B.

MATERIAL and METHODS

This study was performed with a retrospective study design.

Participants

The patients were selected from a referral pool at the Ege Adult Haemophilia and Thrombosis Center (EAHTC). Approval for the study was obtained from the institutional review board at Ege University Ethics Committee. All patients consulted to physiotherapy for routine control between June 1, 2018 and January 30, 2019 were included in the study.

We obtained retrospective case data about physical activity levels and kinesiophobia. The database also comprises existing information about the patient [e.g., age, gender, Body Mass Index (BMI) and hemophilia type]. In most cases, due to the retrospective nature of the event reports, requested data items were missing. A total of 44 patients met the criteria to be included in this study. Individuals were excluded if they had an additional co-morbid illness that may independently affect the outcomes or if they had incomplete data and acute bleeding. Of the 61 patients, 17 had a missing TKS or IPAQ-SF score and were therefore excluded from the study. Records were examined from hemophilic individuals who represented age between 18-60 years.

Assessment parameters

The following information was collected from the database: age, BMI, hemophilia type, kinesiophobia, and

physical activity level assessments data. Kinesiophobia and levels of physical activity were evaluated with a self-reported survey. The data of the questionnaires performed by the patients during their routine control between 1 June 2018 and 30 January 2019 were used for the study.

International Physical Activity Questionnaire- Short Form (IPAQ-SF) was used to evaluate the physical activity levels of participants. (15). The Turkish version of the questionnaire was found valid and reliable (16). This 7-question questionnaire assesses the amount of walking and the amount of moderate and challenging physical activity in work, transportation, housework, gardening and leisure activities performed in the last week. Energy expenditure of queried activities can be calculated for each activity type according to the energy requirements. As a result of the scores obtained, individuals were categorized as high, moderate or low physical activity depending on the total metabolic equivalents in a task (MET)-minute or duration and frequency of each type of physical activity.

Tampa Scale of Kinesiophobia (TSK) was used to assess the fear of movement of the participants. TSK is a self-reported questionnaire that consists of 17-item scored with a 4-point Likert scoring. Total score ranges between 17-68 and higher scores indicate higher levels of kinesiophobia. The reliability and cultural adaptation of the Turkish version was carried out previously (17). The cut-off point of TSK score to determine kinesiophobia was defined as 37 (<37 points indicates a low level of kinesiophobia, while ≥ 37 points indicate a high level of kinesiophobia) (17).

Statistical analysis

The statistical program IBM SPSS Statistics software (release 23.0; IBM Corp., Armonk, NY, USA) was used for the analysis of the data obtained from all variables. As the variables did not have a normal distribution, Spearman correlation analysis was used to measure the degree of association between TSK and IPAQ-SF scores. A p-value of less than 0.05 (typically ≤ 0.05) was referred to a statistically significant difference. The mean \pm standard deviation was used for the descriptive statistic of demographic variables. Percentages and the number of cases were used for the nominal variables.

To compare the differences between TSK and IPAQ-SF scores of patients with hemophilia A and hemophilia B, the Mann-Whitney U test was used.

RESULTS

A total of 44 patients (33 Hemophilia A and 11 Hemophilia B) were included in the study. The average age of the participants was 35.30 ± 12.04 years and mean BMI was 25.68 ± 4.02 kg/m² (Table 1). Of the 44 hemophilic individuals; 33 were with hemophilia A: 20 severe [factor FVIII (FVIII) <0.01 IU mL⁻¹], 9 moderate [FVIII 0.01–0.05 IU mL⁻¹] and 4 mild [FVIII 0.05–0.4 IU mL⁻¹], and 11 were with hemophilia B [4 severe (FIX <0.01 IU mL⁻¹), 5 moderate (FIX 0.01– 0.05 IU mL⁻¹), and 2 mild (FIX 0.05–0.4 IU mL⁻¹)] (Table 2).

High kinesiophobia levels (TSK score of ≥ 37) were found in 90.9 % of patients (40 of 44 patients) (Table 2). On the other hand, when we look at the physical activity levels of patients; 36.4 % had low, 34.1 % moderate and 29.5 % had high physical activity levels (Table 3). There was no statistical correlation between kinesiophobia degree and physical activity level ($p = 0.616$, $r = -0.081$).

Table 1. Demographic characteristics of the participants

Demographic Characteristics	Mean \pm Standard Deviation
Age (years)	35.30 \pm 12.04
Body Mass Index (kg/m ²)	25.68 \pm 4.02

Table 2. Demographic characteristics of the participants

Variable	TSK			Total n (%)
	High kinesiophobia	Low kinesiophobia		
Hemophilia A	30	3		33 (% 75)
Hemophilia B	10	1		11 (% 25)
Total - n (%)	40 (% 90.9)	4 (% 9.1)		44 (%100)
	Disease Severity			
	Severe	Moderate	Mild	Total- n (%)
Hemophilia A	20	9	4	33 (% 75)
Hemophilia B	4	5	2	11 (% 25)
Total - n (%)	24	14	6	44 (%100)

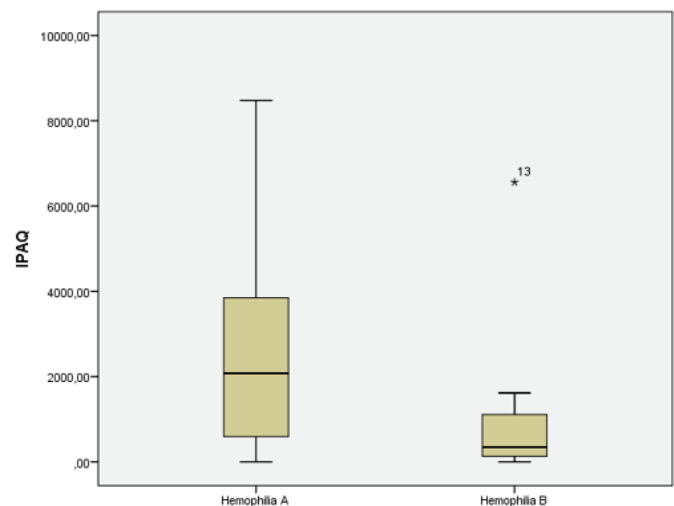
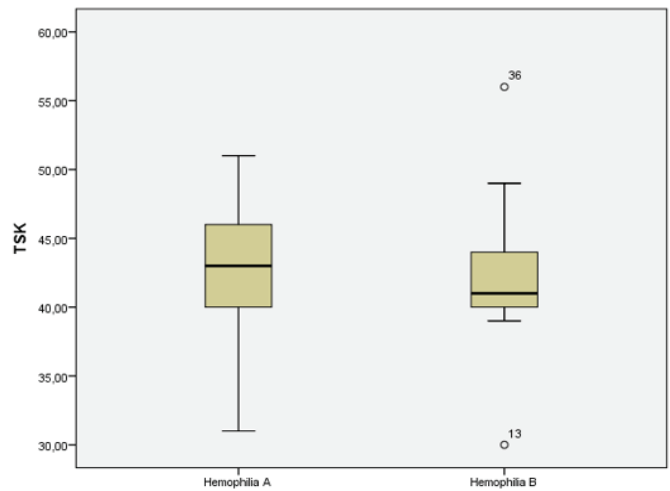
Abbreviations: n: number, TSK: Tampa Scale of Kinesiophobia

Table 3. Distribution of participants according to hemophilia severity and physical activity scores

Variable	Severity of Hemophilia			
	Mild (n)	Moderate (n)	Severe (n)	Total n (%)
Low level activity (n)	1	8	7	16 (36,4%)
Moderate level activity (n)	4	4	7	15 (34,1%)
High level activity (n)	1	2	10	13 (29,5%)
Total- n (%)	6 (13,6%)	14 (31,9%)	24 (54,5%)	44 (100%)

Abbreviations: n: number, IPAQ-SF: International Physical Activity Questionnaire- Short Form

No significant difference was found in kinesiophobia scores between hemophilia A and hemophilia B ($z=-0.707$, $p=0.479$). Physical activity scores differ significantly according to hemophilia type variable in favor of hemophilia A ($z=-2.400$, $p = 0.016$) (Figure 1).



Variables	p	z
TSK	0.479	-0.707
IPAQ-SF	0.016*	-2.400

Abbreviations: TSK: Tampa Scale of Kinesiophobia, IPAQ-SF: International Physical Activity Questionnaire-Short Form

Figure 1. Kinesiophobia and physical activity scores for hemophilia A and hemophilia B

DISCUSSION

This investigation aimed to clarify the relationship between the level of physical activity and kinesiophobia in patients with hemophilia. In our study, we found no correlation between physical activity level and kinesiophobia in patients with hemophilia. However, physical activity levels were different in favor of hemophilia A. These results may be because hemophilia is a chronic disease and hemophilia A is more common than hemophilia B.

The current finding is consistent with evidence from Demirbüken et al. (18) that demonstrated no correlations between physical activity levels and kinesiophobia of people with chronic neck pain. Another study conducted with patients with chronic nonspecific low back pain also reported patients with higher levels of kinesiophobia did not show lower levels of physical activity (9). In contrast to our study Elfving et al. (19) found low levels of physical activity were significantly related to higher scores for TSK in non-specific chronic low back pain patients. Larsson et al. (20) found low kinesiophobia is related to higher levels of physical activity in patients suffering from chronic pain. They suggested that it is important to consider fear-avoidance beliefs in clinical and research to increase physical activity. Contrary to popular belief, this present study found no correlation between the level of physical activity and kinesiophobia in hemophilic individuals. This may be since hemophilia is a lifelong disease. Moreover, the kinesiophobia rates of the included participants were significantly high.

As reported in a review, a high kinesiophobia score is associated with disability, pain, and quality of life in chronic musculoskeletal pain patients (10). People with chronic musculoskeletal pain are reported to reduce physical activity and expected to develop kinesiophobia (10). Symptoms of hemophilia are similar to chronic musculoskeletal pain due to the high percentage of musculoskeletal bleedings. Hence, hemophilic individuals also presumed to develop kinesiophobia and physical inactivity.

The presence of kinesiophobia was high in most of the previous studies on kinesiophobia (21–23). According to the results of our study, 90.9 % of hemophilia patients had high kinesiophobia levels. This high kinesiophobia rate of hemophilia patients suggested that it is important to determine kinesiophobia-related symptoms and the disease-specific cut-off points of kinesiophobia. Furthermore, decreasing the fear of movement should be one goal of hemophilia management and rehabilitation.

In the present study, our findings suggest there was no significant difference between kinesiophobia scores of hemophilia A and hemophilia B. To our knowledge this was the first study to assess the kinesiophobia of hemophilic individuals.

Our findings showed that 36.4 % of patients had low, 34.1 % had moderate and 29.5 % had high physical activity levels. Goto et al. reported significantly lower physical activity levels in people with hemophilia than the recommended level by the WHO (7). Another study reported high physical activity levels (48%) in Irish people with hemophilia (14). However, a few studies reported decreased physical activity levels with increasing age in hemophilia patients (14,24).

Buxbaum et al. suggested similar physical activity levels in boys with hemophilia than healthy boys (ages 11-18) (11).

We compare the physical activity levels of hemophilia A and hemophilia B, because of the lack of a control group. Our results revealed that the clear difference between physical activity levels of hemophilia A and hemophilia B. This difference may be due to hemophilia A is more common than hemophilia B. Patients with hemophilia A had higher physical activity level than those with hemophilia B. Niu et al. (25) reported reduction in time spent on physical activity with age, and a significant decrease in physical activity participation in adults with hemophilia B aged between 25 and 34 years. Given very few studies in this area, these results are considered consistent with our results.

Study Limitations

A limitation of this study was the retrospective design and small sample size that included all levels of severity of hemophilia. A wide range of age in the sample size was also one of the limitations of the study. Therefore there may be differences in physical activity levels. The main limitation of this study was the lack of a control group. Besides, only self-reported measurement data were used in this study. With the use of more objective measures, slightly different results would be achieved. However, these factors were not considered in this study.

CONCLUSION

In conclusion, during the evaluation of hemophilia patients, their attitudes towards activity should be taken into consideration. To our knowledge, this study was the first that investigates the association between kinesiophobia and physical activity levels of patients with hemophilia. Patients with hemophilia should be educated about safe and fearless physical activity. This study also emphasizes quite high the fear of movement in hemophilic individuals. Therefore, there is a need for more detailed studies on their attitudes and behaviors towards movement and physical activity. Further studies are required to entirely understand the role of fear of movement and its interaction with participation in physical activity.

Competing interests: The authors declare that they have no competing interest.

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Ethical approval: The patients were selected from a referral pool at the Ege Adult Haemophilia and Thrombosis Center (EAHTC). Approval for the study was obtained from the institutional review board at Ege University Ethics Committee. All patients consulted to physiotherapy for routine control between June 1, 2018 and January 30, 2019 were included in the study.

Sercan Aykar ORCID: 0000-0002-2843-077X

Filiz Can ORCID: 0000-0003-0641-9956

Fahri Sahin ORCID: 0000-0001-9315-8891

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