

# Kinesiophobia And Related Factors In Adult Patients With Familial Mediterranean Fever

## Kinesiophobie und damit zusammenhängende Faktoren bei erwachsenen Patienten mit familiärem Mittelmeerfieber

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### ABSTRACT

**Objective** Kinesiophobia is a common problem in patients with rheumatic diseases and can cause physical inactivity, social isolation, disability, and poor quality of life. This study aimed to evaluate kinesiophobia and associated factors in patients with familial Mediterranean fever (FMF).

**Methods** A total of 38 patients diagnosed with FMF volunteered to participate in the study. All patients were assessed using the Tampa Kinesiophobia Scale (TKS), the International Physical Activity Questionnaire (IPAQ), the Fatigue Severity Scale (FSS), and the Hospital Anxiety and Depression Scale (HADS).

**Results** Thirty-three (86.8%) of the patients had TKS scores over 37, indicating high levels of kinesiophobia. The TKS score was positively correlated with the HADS depression score ( $r=0.530$ ;  $p=0.001$ ) and the FSS score ( $r=0.340$ ;  $p=0.035$ ) but was not significantly associated with age ( $r=0.102$ ;  $p=0.543$ ), disease duration ( $r=-0.110$ ;  $p=0.511$ ), body mass index ( $r=0.283$ ;  $p=0.085$ ), the HADS anxiety score ( $r=0.306$ ;  $p=0.061$ ), or the IPAQ score ( $r=-0.097$ ;  $p=0.563$ ).

**Conclusions** Our sample of adult FMF patients showed high levels of kinesiophobia associated with fatigue and depression. Treatments focusing on kinesiophobia in FMF patients could help to increase the effectiveness of rehabilitation.

## Introduction

Familial Mediterranean fever (FMF) is a common autoinflammatory disease among certain ethnic groups living in the Mediterranean basin. FMF was reported to be the most common hereditary auto-

inflammatory disease worldwide, possibly due to increased migration [1]. It is characterized by recurrent episodes of one or more indications like peritonitis, pleuritis, arthritis, and fever that cause pain in the abdomen, chest, joints, and muscles, and requires life-

long treatment. The frequency of episodes varies from once a week to once a year, and patients are usually symptom-free between attacks [2]. Musculoskeletal symptoms and arthritis in FMF manifest as acute episodes of pain and swelling affecting one joint at a time and lasting for 1 to 3 days [3]. Before 1975 that colchicine therapy was not used for the management of FMF, patients generally developed amyloidosis, and more than one-third died before the age of 40 [4–6]. Life expectancy has increased due to early diagnosis and proper treatment, thus increasing the importance of physical activity (PA), psychosocial factors, and quality of life [7].

A lack of PA and predominance of sedentary behavior are independent risk factors for comorbidities in the general population [8]. In many disease groups, patients have lower PA levels due to kinesiophobia [9–12]. Kinesiophobia is defined as an excessive fear of physical movement and activity resulting from painful conditions or injuries [13]. Individuals with kinesiophobia do not engage in activities that they believe will increase their pain. Prolonged avoidance of movement leads to a reduction in motor activities due to physical and psychological factors. Kinesiophobia has been defined in rheumatic diseases such as rheumatoid arthritis (RA), ankylosing spondylitis (AS), systemic lupus erythematosus (SLE), and osteoarthritis [8, 9, 14]. In patients with rheumatic diseases, kinesiophobia is associated with inactivity, sleep disturbances, social isolation, depression, anxiety, and pain, all of which can reduce the quality of life. To the best of our knowledge, there are no previous studies investigating kinesiophobia in individuals with FMF. Therefore, the present study was conducted to evaluate kinesiophobia and associated factors in patients with FMF to facilitate the development of comprehensive interventions. Because in the presence of kinesiophobia, aiming to increase activity by reducing symptoms or performing an appropriate intensity exercise load will often not respond. In this case, it should be planned to reduce and/or eliminate kinesiophobia, which is the underlying problem contributing to physical inactivity and symptom severity, with comprehensive interventions like psychosocial rehabilitation, meditation, breathing control, relaxation exercises, and progressive exercise approaches.

## Methods

The current study was conducted in the Haliç University Faculty of Health Sciences, Department of Physiotherapy and Rehabilitation. After obtaining the approval of the university ethics committee, the authors contacted the president of the national Behçet's Disease and Familial Mediterranean Fever Patients Association, a social association where patients are registered with their diagnostic reports. The president invited members to participate in the study by e-mail. A total of 38 of 129 members who had no episodes in the last 6 months responded and volunteered to complete the online survey. None of the participants reported comorbid neurological, neurodegenerative, or infectious diseases at the time of evaluation.

### Demographic and clinical parameters

Demographic characteristics were noted. Clinical parameters evaluated in this study were disease duration, number of episodes in the last year, episode type, and joint involvement.

## Assessments

Levels of fatigue, kinesiophobia, PA, anxiety, and depression were evaluated for all participants. Fatigue severity was assessed using the self-report Fatigue Severity Scale (FSS). The FSS assesses the effect of fatigue on daily life and consists of 9-item. Each item is scored from 1 (completely disagree) to 7 (completely agree). FSS score is determined by calculating the average of the 9 items. A higher score indicates more severe fatigue. The cut-off value for pathological fatigue was determined as 4 and above [15].

Kinesiophobia was evaluated using the Tampa Kinesiophobia Scale (TKS). The TKS consists of 17 items scored on a 4-point likert scale and measures fear of movement/(re)injury. The total score is obtained by summing all items, and a higher score indicates greater fear of movement. A total score greater than 37 is regarded as indicating a high level of kinesiophobia [13].

The Hospital Anxiety and Depression Scale (HADS) is a widely used and reliable self-assessment tool for determining depression and anxiety states. The aim of the scale is not to diagnose, but to evaluate patients' psychological states to take the necessary actions [16]. The scale consists of 14 items divided into the depression and anxiety subscales, each with 7 items. The items on the scale are scored from 0 to 3 using a 4-point likert scale. The scores of the odd-numbered items are summed to obtain the anxiety subscale (HADS-A) score and the even-numbered items are summed to obtain the depression subscale (HADS-D) score. Possible scores for each subscale range from 0 to 21. In the study conducted with the Turkish version of the scale, cut-off scores of 10 and 7 were identified for the anxiety and depression subscales, respectively [17].

The short form of the International Physical Activity Questionnaire (IPAQ) was used to assess PA [18]. In the IPAQ, respondents estimate how often and for how long they engaged in various activities in the last 7 days. PA levels were classified into 3 categories: inactive (< 600 MET (metabolic equivalent) -min/week), low PA level (600–3000 MET-min/week), and adequate PA level (> 3000 MET-min/week) [18].

### Statistical analyses

The G-power version 3.1 program (Universitaet Kiel, Germany) was used to determine the sample size [19]. For 95% power, 0.58 effect size, and 95% confidence interval, the necessary sample size was calculated as 28 [14]. Statistical analysis was performed using IBM SPSS Statistics version 22.0 (IBM Corp., Armonk, NY). Normality of data distributions was determined using the Kolmogorov–Smirnov test. The data were expressed as mean  $\pm$  standard deviation (SD) for quantitative variables and as a percentage (%) for categorical variables. Correlations between variables were estimated with the Spearman correlation coefficient.  $p < 0.05$  was considered statistically significant for all analyses.

## Results

Thirty-eight FMF patients (23 female, 15 male) with a mean age of  $37.05 \pm 8.71$  years were enrolled in the study. Patients' demographic and clinical characteristics including age, disease duration, body mass index, and questionnaire results, are presented in ► **Table 1**. Thirty-three (86.8%) of the subjects had a TKS score over 37, reflect-

► **Table 1** Patients' demographic and clinical data.

	Mean ± SD	Min.	Max.
Age (years)	37.05 ± 8.71	19	57
Disease duration (years)	13.11 ± 11.14	1	46
BMI (kg/cm <sup>2</sup> )	24.5 ± 4.43	16.65	38.05
IPAQ	6942.5 ± 2520.4	3018	14580
TKS	44.16 ± 5.06	33	54
HADS-A	9.84 ± 4.43	2	20
HADS-D	8.82 ± 4.70	1	20
FSS	56.45 ± 13.05	23	70

SD: Standard deviation; Min: Minimum; Max: Maximum; BMI: Body Mass Index; IPAQ: International Physical Activity Questionnaire; TKS: Tampa Kinesiophobia Scale; HADS-A: Hospital Anxiety and Depression Scale – anxiety subscale; HADS-D: Hospital Anxiety and Depression Scale – depression subscale; FSS: Fatigue Severity Scale.

► **Table 2** Correlations between Tampa Kinesiophobia Scale scores and other parameters.

	r-value	p-value
Age (years)	0.102	0.543
Disease duration (years)	-0.110	0.511
BMI (kg/cm <sup>2</sup> )	0.283	0.085
IPAQ	-0.097	0.563
HADS-A	0.306	0.061
HADS-D	0.529	<b>0.001</b>
FSS	0.344	<b>0.035</b>

BMI: Body Mass Index; IPAQ: International Physical Activity Questionnaire; HADS-A: Hospital Anxiety and Depression Scale – anxiety subscale; HADS-D: Hospital Anxiety and Depression Scale – depression subscale; FSS: Fatigue Severity Scale.; \* Significance evaluated at the 0.05 level; significant results are shown in bold.

ing high kinesiophobia. The mean IPAQ score was 6942.5 ± 2520.4, indicating sufficient PA. The mean HADS-A score was 9.84 ± 4.43, which is below the cut-off value, while the mean HADS-D score was 8.82 ± 4.70, which is higher than the cut-off value. The mean FSS score was 56.45 ± 13.05, indicating a high level of fatigue.

TKS scores were positively correlated with HADS-D ( $r = 0.530$ ;  $p = 0.001$ ) and FSS ( $r = 0.340$ ;  $p = 0.035$ ) but showed no significant correlation with age ( $r = 0.102$ ;  $p = 0.543$ ), disease duration ( $r = -0.110$ ;  $p = 0.511$ ), body mass index ( $r = 0.283$ ;  $p = 0.085$ ), HADS-A score ( $r = 0.306$ ;  $p = 0.061$ ), or IPAQ score ( $r = -0.097$ ;  $p = 0.563$ ) (► **Table 2**). Gender and regular exercise did not affect TKS scores ( $p > 0.05$ ) (► **Table 3**).

► **Table 3** Comparison of the patients' TKS scores according to age gender and regular exercise.

		N	TKS Score Mean ± SD	p-value*
Gender	Male	15	44.07 ± 4.3	0.930
	Female	23	44.22 ± 5.59	
Regular Exercise	Yes	9	43.11 ± 4.88	0.485
	No	29	44.48 ± 5.15	

TKS: Tampa Kinesiophobia Scale; SD: Standard deviation; \* Significance evaluated at the 0.05 level.

## Discussion

In this study, the sample of adult subjects with FMF had high levels of kinesiophobia which was associated with fatigue and depression. Age, gender, body mass index, disease duration, PA level, and anxiety were not related to kinesiophobia. To the best of our knowledge, this is the first study examining kinesiophobia as a symptom in FMF patients.

Kinesiophobia is an important issue that decreases exercise capacity, increases symptoms like muscle weakness, pain, and fatigue, and affects the quality of life in various patient groups. Studies on the presence of kinesiophobia in rheumatic diseases are steadily increasing in number and generally report high levels of kinesiophobia among these patients [9, 14, 20]. Rates of high kinesiophobia were reported in 66 % of SLE patients and 60.6 % of RA patients [9, 20]. In our study, 86 % of the patients had TKS scores over 37, representing high kinesiophobia.

Fatigue is a symptom that affects both physical and cognitive functions in people with rheumatic disease. In a study of FMF patients by Duruoz et al., the median FSS score was 44, whereas the mean FSS score was 56.45 in our study [7]. For RA patients, it was reported that over 80 % had clinically significant fatigue and over 50 % had severe fatigue [21]. We also determined in current study that FMF patients had a high level of fatigue and it was correlated with kinesiophobia.

According to current results, the mean depression score (8.82 ± 4.70) was higher than the HADS-D cut-off value, while the mean anxiety score (9.84 ± 4.43) was below the HADS-A cut-off value. Studies that assessed depression as a symptom of FMF found that patients had higher scores compared to healthy peers [7, 22, 23]. In addition, the scores reported in those studies were similar to current findings. Duruoz et al. [7] reported HADS-D and HADS-A mean scores of 7 and 9, while Deger et al. [23] reported median scores of 6 and 8, respectively. Due to the absence of a healthy control group, we compared scores with the cut-off values for the normal population. Nevertheless, considering the literature data, current results indicated higher levels of depression and anxiety.

In current patient group, depression scores were associated with kinesiophobia while anxiety scores were not. Correlation with psy-

chosoical factors has also been observed previously in different rheumatoid diseases. Studies in SLE [9] and AS [24] demonstrated a correlation between Beck depression score and kinesiophobia.

FMF is a periodic disease characterized by recurrent self-limiting inflammatory febrile attacks of serositis, arthritis, and erysipelas-like erythema. Mild or moderate episodes impede daily activities to varying degrees depending on the type and severity of the attack. Babaoglu et al. demonstrated that FMF episodes significantly impaired patients' PA when assessed with activity tracking [25]. When we assessed PA using the IPAQ, the results suggested that current patient group had normal PA levels. This may be related to the fact that the FMF patients in current study had not had an acute episode in at least 6 months and were able to completely return to their routine. The IPAQ assesses PA levels in the last 7 days, but patients' physical inactivity likely corresponds to their symptoms. If the patients had been evaluated during the symptomatic period, they may have reported lower levels of PA and a relationship between PA level and kinesiophobia may have been detected. PA levels of the subjects were sufficient and there was no relationship between PA and kinesiophobia. This surprising lack of correlation may be interpreted as patients maintaining their PA level despite having kinesiophobia, as a result of learning and adapting to living with a chronic disease.

Several kinesiophobia-related factors have been identified in different rheumatoid diseases. In the sample of adult FMF patients of current study, kinesiophobia was associated with fatigue and depression. In SLE patients, one study detected a significant relationship between kinesiophobia and depression, while there was no relationship with PA, disease activity, or fatigue [9]. In patients with RA, high fear-avoidance beliefs about PA were associated with male gender, below-average income, high level of pain, poor health, and low health-related quality of life score [20]. In literature, psychological factors like depression are mostly related to kinesiophobia but other related factors were changed according to the type of disease, symptoms, and clinics [26].

To the best of our knowledge the current study is the first study to determine the prevalence of kinesiophobia and associated factors in patients with FMF. On the other hand, the absence of a healthy control group is a limitation. Further controlled studies on this topic are needed. As with other rheumatic diseases, exercise is of great importance in the rehabilitation of FMF patients because of its positive effects on the musculoskeletal system. However, because rheumatic diseases are chronic inflammatory conditions with activation and remission periods, ensuring treatment adherence can present a challenge. Identifying and addressing kinesiophobia in these patients may contribute to their rehabilitation.

## Conclusions

The sample of adult FMF patients in the current study showed high levels of kinesiophobia, which was associated with fatigue and depression. It is predicted that treatment and rehabilitation strategies that consider kinesiophobia in FMF patients will be more beneficial in increasing the success of disease management.

## Conflict of Interest

The authors declare that they have no conflict of interest.

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