DOI: https://doi.org/10.18621/eurj.1292729

Physical Medicine and Rehabilitation

Fibromyalgia syndrome in mothers of children with cerebral palsy and its relationship with caregiver burden: a cross-sectional study

İlknur Aykurt Karlıbel®, Meliha Kasapoğlu Aksoy®, Merve Yeni®

Department of Physical Medicine and Rehabilitation, University of Health Sciences, Bursa Yüksek Ihtisas Training and Research Hospital, Turkey

ABSTRACT

Objectives: Fibromyalgia syndrome (FMS) is a stress-related disease. The birth of a child with cerebral palsy (CP) is an important stress factor for the mother. This study aims to investigate the incidence of FMS in mothers of children with CP by comparing it with the control group and determining the factors affecting the severity of FMS.

Methods: The study included 112 children with CP (age: 38-216 months), their caregiver mother (age: 23-50 years) (Group 1) and 52 non-disabled children (age: 40-180 months), their caregiver mothers (age: 27-50 years) (Group 2). Children were evaluated with the Gross Motor Function Classification System (GMFCS) and the functional independence scale for children (WeeFIM). The mothers were evaluated according to the 2010 ACR FMS diagnostic criteria. The FMS Impact Questionnaire (FIQ), the Hospital Anxiety and Depression Scale (HADS), and the Bakas Caregiving Outcomes Scale (BCOS) were applied.

Results: In Group 1, mothers had higher anxiety-depression scores and caregiving burden than Group 2 (P<0.05). FMS rate was %31.3 in Group 1 and %5.7 in Group 2. Mothers' anxiety-depression scores, widespread pain index, symptom severity score, pain, and caregiver burden were higher in Group 1 than in Group 2 (P<0.05). Factors affecting the severity of FMS are the number of siblings of children with CP, the number of siblings with CP, GMFCS, dependence level, anxiety-depression levels of mothers, and caregiver burden. The most influential factor is the caregiver burden.

Conclusions: Long-term heavy caregiver burden in mothers of children with CP may be effective in developing FMS.

Keywords: Caregiver burden, cerebral palsy, fibromyalgia, mothers, pain

erebral palsy (CP) is one of the most common causes of physical and developmental disability in childhood [1]. Abnormal muscle tone, loss of voluntary motor control, sensory and coordination disorders and muscle weakness are the most

important characteristics of the disease. Cognitive-behavioral abnormalities, speech, vision problems, epilepsy, urinary system problems and nutritional problems may also accompany [2]. This situation brings along the dependency of children with CP on

Corresponding author: İlknur Aykurt Karlıbel, MD., Associate Professor. Phone: +90 224 295 50 00. E-mail: karlibeli@hotmail.com

How to cite this article: Aykurt Karlıbel İ, Kasapoğlu Aksoy M, Yeni M. Fibromyalgia syndrome in mothers of children with cerebral palsy and its relationship with caregiver burden: a cross-sectional study. Eur Res J. 2024;10(1):59-69. doi: 10.18621/eurj.1292729

Received: May 5, 2023 Accepted: August 8, 2023 Published Online: September 9, 2023

Copyright © 2024 by Prusa Medical Publishing Available at http://dergipark.org.tr/eurj





This is an open access article distributed under the terms of Creative CommonAttribution-NonCommercial-NoDerivatives 4.0 International License

their caregivers at different rates in their daily living activities. Mothers bear the most responsibility for caring and often lack carer support [3].

The birth of a disabled child is an important stress factor for the family and mothers are affected the most. Mothers suffer from feelings of guilt about the child's condition. CP requires lifelong care and adversely affects parents' physical and psychological health [4]. Caring for a child with CP creates a financial burden on the family and causes limitations in social and cultural activities. Also, families are exposed to social stigma. Incidents of domestic violence are also common [3, 5]. Studies have shown that caregivers have high anxiety and depression and that caregiving impairs their quality of life [3, 6]. In addition, physical problems such as insomnia, intense physical activityrelated musculoskeletal pain and hypertension have also been reported. The mother cannot find time to rest [3, 7].

Fibromyalgia is a stress-related disorder characterized by widespread chronic pain, profound fatigue, and sleep disturbances. It is counted among the central sensitivity syndromes [8, 9]. In the literature, the prevalence of fibromyalgia has been reported between 0.2% and 6.6% in the general population, and between 2.4% and 6.8% in women [10]. Emotional, physical, and environmental stressful events are frequently observed in patients with fibromyalgia [9]. Studies have shown that depression and anxiety are more common in fibromyalgia patients than healthy individuals, and that the presence of depression and anxiety increases pain severity and worsens quality of life [11]. In addition, studies have shown the relationship between posttraumatic stress disorder and fibromyalgia [9, 12]. In a study where the majority of the participants were women, 90% of the participants exhibited somatic symptom disorder. It is claimed that fibromyalgia patients may have neuropathic pain triggered by stress rather than a mental somatic symptom disorder. This is explained by the stress-response system dysfunction in fibromyalgia patients and the presence of abnormal connections between the stress response system and pain-transmitting nerves within the dorsal root ganglia [9].

In the light of this information, we thought that the emotional, physical and environmental chronic stress created by the burden of care for the child with CP may increase the incidence of fibromyalgia in mothers who often have to undertake the responsibility of care

alone. However, according to our research, we could not find a study in this direction.

Objectives of our study: (1) To investigate the incidence of fibromyalgia in mothers of children with CP in comparison with the control group, (2) Comparison of mothers with and without fibromyalgia in terms of caregiving burden, depression, anxiety levels, and "children's functional status and independence levels," (3) To determine the factors affecting the severity of fibromyalgia in mothers of children with CP.

METHODS

The cross-sectional, observational clinical study was conducted in a training and research hospital's physical medicine and rehabilitation clinics (from May 2019 to March 2020). The study was planned under the Helsinki Declaration rules, and local ethics committee approval was received (2011-KAEK-25 2019 / 04-03). All participants were informed about the study and signed a written consent form.

Study Population

In this study, the study group consisted of mothers who cared for a child diagnosed with Cerebral Palsy aged 3 to 18 years (Group 1; n=140). The control group consisted of mothers of children aged 3 to 18 years who applied to the clinic with other diagnoses (Group 2; n=60).

Exclusion criteria: (1) not having sufficient cognitive function to evaluate the questionnaire questions, (2) having diseases such as infection, chronic systemic disease (cardiac, renal, endocrine diseases, etc.), rheumatological disease, neurological disease, psychotic disease, malignancy, etc., (3) pregnancy and breastfeeding.

In addition, children whose caregivers were not mothers were also excluded from the evaluation.

Intervention

The demographic data (age, body mass index [BMI kg/m²] of children with CP, children in the control group, and their mothers were recorded.

Children

A physiatrist examined children with cerebral palsy, and the diagnosis was confirmed based on med-

ical records. CP type was determined (spastic diplegic, spastic quadriplegic, spastic hemiplegic, dyskinetic, ataxic) [13].

The Gross Motor Function Classification System (GMFCS) was used to evaluate the motor development of children with CP [14]. GMFCS assesses the gross motor development of children with CP at five levels: (1) They walk without difficulty. There is difficulty in more advanced gross motor skills. (2) They walk without assistive devices. There are restrictions on walking outside the home and in the community. (3) They walk with mobility aids. There are restrictions on walking outside the home and in the community. (4) Self-movement is limited. Children are carried on laps or use powered mobility devices outside the home and in the community is severely limited, even with the use of assistive technology.

The functional independence scale for children (WeeFIM) [15] was used to assess the functional status of children with CP. WeeFIM is examined under three subheadings: self-care, mobility, and cognitive function. A total of 18 questions are scored between 1 and 7. The lowest score is 18, and the highest score is 126.

Mothers

The mothers of the children were evaluated according to 2010 the American College of Rheumatology (ACR) FMS diagnostic criteria [16]. Mothers were questioned for pain at 19 specific body points. Sensitivity for FMS was assessed by applying a pressure of 4 kg/cm² to 19 specific body points. Thus, the patients' widespread pain index (WPI) was determined. Mothers were also questioned in terms of fatigue, waking unrefreshed, cognitive symptoms, and somatic symptoms according to 2010 ACR FMS diagnostic criteria, and symptom severity score (SSS) was determined. Mothers who defined generalized pain for more than three months were diagnosed with FMS according to the 2010 ACR FMS diagnostic criteria if the WPI was ≥ 7 , SSS was ≥ 5 , or WPI was 4-6, SSS was ≥ 9 .

The pain intensity of the patients was evaluated using a visual analog scale (VAS) (On a 10 cm analog scale, 0 means no pain, ten means unbearable pain.), and functional status was evaluated using the FMS Impact Questionnaire (FIQ).

The FIQ is a valid and reliable method for evalu-

ating the impact of FMS on daily life [17]. It measures ten characteristics: physical function, feeling unwell, absenteeism, difficulty at work, pain, fatigue, morning fatigue, stiffness, anxiety, and depression. The maximum score for the FIQ is 100, with higher scores indicating higher disease severity. A total FIQ score of 0-<39 indicates low impact, \ge 9<59 indicates moderate impact, and \ge 59-100 indicates severe impact.

Mothers' anxiety and depression levels were evaluated using the Hospital Anxiety and Depression Scale (HAD). The scale consists of 14 questions, with each question being scored between 0-3. Seven questions (the odd-numbered items) measure anxiety (HAD-A), and the other seven (evenly numbered) measure depression (HAD-D). The score range for each anxiety and depression subscale is 0-21. 0-7 points indicate normal levels, 8-10 points indicate borderline abnormal levels, and 11-21 points indicate abnormal levels [18].

Mothers' caregiving burdens were evaluated with the Bakas Caregiving Outcomes Scale (BCOS) [19]. This scale was used to evaluate how the caregivers' lives changed with the start of the caregiving process. The scale shows both positive and negative effects. The scale consists of 15 questions scored between +3 (in the best direction) and -3 (in the worst direction). It is a Likert-type scale ranging from 1 to 7. A minimum of 15 and a maximum of 105 points can be obtained from the scale.

First, mothers of children with CP and mothers of children in the control group were compared in terms of evaluation parameters, and FMS rates of both groups were determined. The evaluation parameters of mothers of children with CP with and without FMS were compared in the next step. Factors affecting FMS severity were determined.

Statistical Analysis

IBM SPSS 23.0 statistical software was used to analyze the data. Descriptive statistical methods (frequency, percentage, median, and min-max) were used while evaluating the study's data. The data's compliance with a normal distribution was assessed using Shapiro-Wilk tests. Where the data showed normal distribution, a t-test was used. Otherwise, Wilcoxon Test and Mann-Whitney U test were used to make intergroup comparisons. A Chi-square test was used to compare categorical data. The relationships between variables were assessed using Spearman's rho corre-

lation test. Univariate and multivariate logistic regression analyses investigated the factors affecting FMS severity.

RESULTS

The study included one hundred twelve children with CP and their caregiver mothers (Group 1). The median age of the children was 106.5 months (range: 38-216 months), and the median age of the mothers was 35 years (range: 23-50 years). In the control group, 52 children and their mothers were included in the study (Group 2) (Fig. 1). The median age of the children was

108 months (range: 40-180 months), and the median age of the mothers was 34 years (range: 27-50 yeras).

In Table 1, the results of the evaluation parameters of the mothers of children with CP and the mothers in the control group are given: There was no statistically significant difference between the two groups in terms of demographic data. Mothers of children with CP had statistically significantly higher scores in anxiety and depression scores, WPI, SSS, and pain severity (VAS) than mothers in the control group (P<0.001). Mothers of children with CP had statistically significantly lower scores than mothers in the control group regarding caregiving burden (P<0.001). Mothers of children with CP received statistically significantly more care-

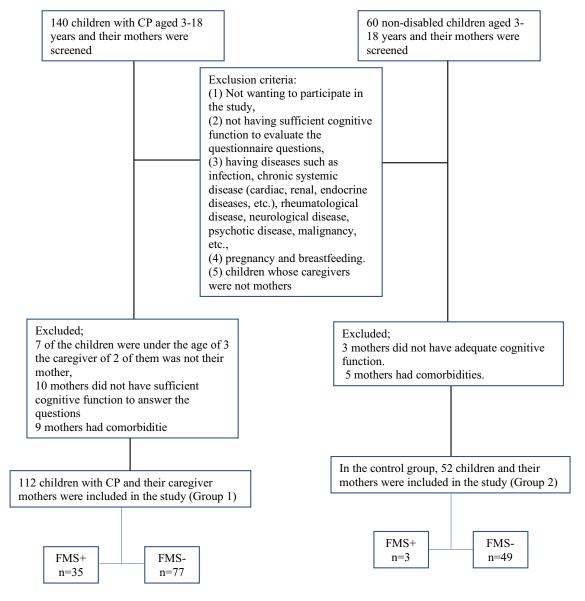


Fig. 1. Flow chart.

giver support than the control group (P<0.001). FMS was detected in 31.3% of Group 1. The FMS rate of Group 2 was 5.7%.

Demographic characteristics and clinical and functional status of children with CP and children in the control group are given in Table 2: The BMI (kg/m^2) values of the children with CP were significantly lower than the children in the control group (P<0.001).

The comparison of mothers of children with CP with and without FMS in terms of evaluation parameters is given in Table 3: Mothers with FMS of chil-

dren with CP exhibited statistically significantly higher anxiety and depression scores; perceived caregiver burden was also statistically significantly higher. Children with CP of mothers with FMS had statistically significantly older, lower BMI, and more siblings and siblings with CP than those without FMS. Children with CP of mothers with FMS had a statistically significant higher GMFCS score and lower WeeFIM sub-parameters and total scores compared to those without FMS.

Table 4 shows the relationships between maternal FMS severity, pain severity, psychological status, care-

Table 1. Demographic characteristics of the mothers of children with CP, FMS, psychological status assessment, and caregiving burden

		Mothers of children Control group with CP		P value	
		(n=112)	(n=2)		
Age (year)		35 (23-50)	34 (27-50)	0.897	
BMI (kg/m ²)		25.2 (18.8-37.3)	26.6 (19.7-32.4)	0.192	
Marital status, n (%)	Married	103 (92%)	50 (94.3%)	0.584	
	Single	9 (8%)	3 (5.7%)		
Working status, n (%)	Working	13 (11.6%)	8 (15.1%)	0.532	
	Not working	99 (88.4%)	45 (84.9%)		
Social insurance, n (%)		110 (98.2%)	53 (100%)	0.329	
Perceived income, n (%)	Income < expenses	54 (48.2%)	30 (56.6%)	0.260	
	Income = expenses	47 (42%)	20 (37.7%)		
	Income > expenses	11 (9.8%)	3 (5.7%)		
Education n (%)	Primary education	65 (58%)	27 (50.9%)	0.450	
	High school	33 (29.5%)	19 (35.8%)		
	University	14 (12.5%)	7 (13.2%)		
Caregiver assistance n (%)		38 (33.9%)	2 (3.8%)	<0.001	
HADS-A		11 (4-21)	6 (4-13)	<0.001	
HADS-D		12 (5-20)	7 (4-13)	<0.001	
BCOS		53.50 (25-65)	66 (53-76)	<0.001	
WPI		1 (0-12)	0 (0-8)	<0.001	
SSS		5 (2-12)	3 (2-6)	< 0.001	
Pain (VAS)		4 (0-10)	0 (0-7)	<0.001	
FMS (+), n (%)		35 (31.3%)	3 (5.7%)	<0.001	

Data are shown as median (minimum-maximum) or n (%). CP=Cerebral palsy, FMS=Fibromyalgia syndrome, HADS-A=Hospital anxiety scale, HADS-D=Hospital depression scale, VAS=Visual analog scale, FIQ=FMS Impact Questionnaire, BCOS=Bakas Caregiving Outcomes scale, WPI=Widespread pain index, SSS=symptom severity scale, VAS=Visual analog scale

giving burden, and functional status of the child with CP: There was a statistically significant positive correlation between the FIQ scores and the total number of siblings of the children with CP, the number of siblings with CP, the GMFCS score, and the anxiety and depression scores of their mothers (P<0.05). There was a negative correlation between FIQ scores and WeeFIM self-care, mobility, cognitive function subscores, WeeFIM total score, and BCOS scores of children with CP (P<0.05).

Univariate and multivariate logistic regression analyses were performed to determine the factors affecting fibromyalgia severity (Table 5). In the multivariate analysis, only four of the ten variables with statistically significant odds ratios (OR) in the univariate regression analysis remained statistically signifi-

cant. Outcomes that had statistical significance in Univariate regression analysis were number of siblings, number of siblings with CP, GMFCS level, Wee-FIM self-care, Wee-FIM mobility, Wee-FIM cognitive functions, Wee-FIM total, maternal anxiety and depression levels, and mother's caregiver burden. Outcomes that had statistical significance in the multivariate analysis were (in order of importance): mother's caregiver burden, mother's depression levels, GMFCS level of the child with CP, and a number of siblings with CP.

DISCUSSION

Our study showed that mothers of children with CP

Table 2. Demographic characteristics, clinical and functional status of children with CP and control group

		Children with CP (n=112)	Control group (n=52)	P value
Age (month)		106.5 (38-216)	108 (40-180)	0.536
Gender	Female	60 (53.6%)	35 (66%)	0.131
	Male	52 (46.4%)	18 (34%)	
BMI (kg/m^2)		17.20 (14.10-20.80)	18.90 (16.50-22)	< 0.001
Number of siblings		1 (0-5)	1 (0-3)	0.875
Siblings with CP		11 (9.8%)	-	
Clinical type of SP n (%)	Spastic diplegia	29 (25.9%)	-	
	Spastic quadriplegia	44 (39.3%)	-	
	Spastic hemiplegia	32 (28.6%)	-	
	Dyskinetic & Ataxic	7 (6.3%)	-	
GMFCS n (%)	GMFCS 1	14 (12.5%)	-	
	GMFCS 2	43 (38.4%)	-	
	GMFCS 3	21 (18.8%)	-	
	GMFCS 4	18 (16.1%)	-	
	GMFCS 5	16 (14.3%)	-	
GMFCS, Median (min-max	x)	2 (1-5)		
WeeFIM self-care		24 (5-42)	-	
WeeFIM mobility		20.50 (5-35)	-	
WeeFIM cognitive function	n	28 (5-35)	-	
WeeFIM total		83.50 (18-126)	-	

Data are shown as median (minimum-maximum) or n (%). CP=Cerebral palsy, GMFCS=Gross Motor Function Classification System, WeeFIM=Functional independence scale for children

Tablo 3. Comparison of mothers with and without FMS of children with CP

		FMS (+) (n=35)	FMS (-) (n=77)	P value
Mothers of children with	CD	(n-33)	(n-//)	
Age (year)		37 (26-50)	34 (23-49)	0.121
BMI (kg/m ²)		27.10 (18.80-37.30)	24.70 (19.70-32)	0.121
Marital status n (%)	Married	31 (88.5%)	71 (92.2%)	0.130
Marital Status II (%)		` ′		0.373
Walling status a (0/)	Single	4 (11.5%)	6 (7.8%)	0.069
Working status n (%)	Yes	4 (11.5%)	8 (10.3%)	0.968
Education of (0/)	No Drive a re-	31 (88.5%)	69 (89.7%)	0.062
Education n (%)	Primary	25 (71.4%)	40 (52.0%)	0.062
	High school	7 (20%)	25 (32.4%)	
D . 1. (0/)	University	3 (8.6%)	12 (15.6%)	0.200
Perceived income n (%)	Income <expenditure< td=""><td>19 (54.3%)</td><td>38 (49.3%)</td><td>0.300</td></expenditure<>	19 (54.3%)	38 (49.3%)	0.300
	Income=expenditure	14 (40%)	33 (42.8%)	
	Income>expenditure	2 (5.7%)	6 (7.9%)	
HADS-A		12 (6-21)	11 (4-17)	<0.001
HADS-D		15 (9-20)	11 (5-20)	< 0.001
Pain (VAS)		7 (4-10)	-	
FIQ		69.99 (60.32-94.26)	-	
BCOS		47 (25-60)	55 (38-65)	< 0.001
Children with CP				
Age (month)		117 (41-216)	99 (38-214)	0.038
Gender	Female	18 (51.4%)	42 (54.5%)	0.760
	Male	17 (48.6%)	35 (45.5%)	
BMI (kg/m^2)		16.60 (14.10-19.90)	17.60 (14.10-20.80)	0.004
Number of siblings		1 (0-3)	1 (0-5)	0.006
Siblings with CP n (%)		8 (22.8%)	3 (3.8%)	0.002
Recipients of caregiver ass	sistance	11 (31.4%)	27 (35.1 %)	0.708
Clinical type of Cp n (%)	Spastic diplegia	8 (22.9%)	21 (27.3%)	0.286
	Spastic quadriplegia	21 (60%)	23 (29.9%)	
	Spastic hemiplegia	2 (5.7%)	30 (39%)	
	Dyskinetic & Ataxic	4 (11.4%)	3 (3.9%)	
GMFCS n (%)	GMFCS 1	1 (2.9%)	13 (16.9%)	< 0.001
	GMFCS 2	5 (14.3%)	38 (49.4%)	
	GMFCS 3	4 (11.4%)	17 (22.1%)	
	GMFCS 4	13 (37.1%)	5 (6.5%)	
	GMFCS 5	12 (34.3%)	4 (5.2%)	
WeeFIM self-care		12 (5-37)	27 (6-42)	< 0.001
WeeFIM mobility		10 (5-35)	25 (5-35)	< 0.001
WeeFIM cognitive function	on	17 (5-35)	29 (5-35)	0.001
WeeFIM total		57 (18-110)	93 (18-126)	< 0.001

Data are shown as median (minimum-maximum) or n (%). CP=Cerebral palsy, FMS=Fibromyalgia syndrome, HADS-A=Hospital anxiety scale, HADS-D=Hospital depression scale, VAS=Visual analog scale, FIQ=FMS Impact Questionnaire, BCOS=Bakas Caregiving Outcomes scale, GMFCS=Gross Motor Function Classification System, WeeFIM=Functional independence scale for children

Table 4. The relationship between maternal FMS severity, psychological status, functional status, dependency level of the child with CP, and caregiving burden

	F	'IQ
	R	P value
Children with CP		
Age (year)	0.168	0.077
Gender	0.013	0.893
Number of siblings	0.249	0.008
Siblings with CP	0.300	<0.001
Clinical type of CP	-0.106	0.268
GMFCS	0.555	< 0.001
WeeFIM self-care	-0.468	<0.001
WeeFIM mobility	-0.426	< 0.001
WeeFIM cognitive function	-0.360	<0.001
WeeFIM total	-0.482	<0.001
Mothers of children with CP		
Recipients of caregiver assistance	-0.610	0.523
HADS-A	0.360	<0.001
HADS-D	0.532	<0.001
BCOS	-0.589	<0.001

CP=Cerebral palsy, FMS=Fibromyalgia syndrome, HADS-A=Hospital anxiety scale, HADS-D=Hospital depression scale, VAS=Visual analog scale, FIQ=FMS Impact Questionnaire, BCOS=Bakas Caregiving Outcomes scale, GMFCS=Gross Motor Function Classification System, WeeFIM=Functional independence scale for children

had higher levels of anxiety, depression, and caregiving burden and higher FMS rates than the control group. Almost a quarter of mothers of children with CP had FMS. It was found that mothers with FMS of children with CP had higher anxiety, depression, and caregiving burden than those without FMS. Children with CP of mothers with FMS had a higher number of siblings and siblings with CP, advanced age, lower motor functions, and higher dependency levels than those without FMS. This study determined that the factors affecting FMS severity were the number of siblings of children with CP, the number of siblings with CP, GMFCS level, dependency level, mothers' anxiety and depression levels, and mother's caregiver burden. The most influential factor in FMS severity was the

mother's caregiver burden. This was followed in order of importance by the mother's depression, the GMFCS level, and the number of the sibling with CP.

CP is the most disabling disease of childhood and requires lifelong care. In this process, parents' physical and psychological health is adversely affected [1, 4]. Previous studies have reported that musculoskeletal pain is more common in mothers of children with cerebral palsy than in mothers of healthy children [20, 21]. More commonly, spinal pain has been investigated [20-22]. In a study evaluating back pain and underlying causes in mothers of children with CP, only 20.67% of mothers did not complain of pain [22]. In the literature, the prevalence of FMS in women has been reported as 2.4% - 6.8% [10]. In the current study, 31.3% of mothers of children with CP were diagnosed with FMS. This rate was significantly different from the control group.

It has been found that the incidence and intensity of musculoskeletal pain in mothers of children with CP primarily depend on the child's functional status and dependency level, body weight, age, and intense physical activity load created by caregiving [3, 22]. In contrast, Byrne et al. [23] reported that CP severity did not affect parents' health status (including musculoskeletal pain). In addition, having more than one child with CP and maternal depression levels have been identified as risk factors for musculoskeletal pain in mothers [20]. Caring for a child with CP is a oneway, dependent, and long-term process. Therefore, as the child's age increases, the caregiver burden increases [20, 24]. In the current study, children with CP of mothers with FMS were older and had more siblings and siblings with CP, consistent with the literature. Contrary to the literature, the children of mothers with FMS had lower BMI. The developmental delay in the child may have caused the mothers to feel inadequate about feeding. Feelings of inadequacy have been significantly associated with anxiety and depression [25].

The quality of life of mothers of children with CP is adversely affected by various factors such as caregiver burden, fatigue, and psychological symptoms [26]. There are studies stating that the severity of CP negatively affects the mother's quality of life, as well as studies that do not support this [21, 23]. Caregivers of children with cerebral palsy suffer from a significant psychosocial burden, and this burden was found

Tablo 5. Evaluation of the factors affecting the severity of FMS in mothers of children with CP

	Univariate analysis			Multivariate analysis				
	95% CI for OR			95% CI for OR				
	OR	Lower	Upper	P value	OR	Lower	Upper	p value
Children with CP								
Age	1.009	0.999	1.018	0.073				
Gender	1.372	0.648	2.903	0.408				
Number of siblings	1.591	1.098	2.305	0.014				
Siblings with CP	8.585	2.459	269.966	0.001	6.974	1.108	43.886	0.039
Recipients of caregiver assistance	1.596	0.700	3.641	0.266				
Clinical type of CP	0.835	0.526	1.324	0.443				
GMFCS	3.171	2.046	4.916	< 0.001	3.169	1.568	6.406	0.001
WeeFIM self-care	0.936	0.909	0.964	< 0.001				
WeeFIM mobility	0.902	0.859	0.947	< 0.001				
WeeFIM cognitive function	0.935	0.900	0.971	< 0.001				
WeeFIM total	0.970	0.956	0.983	< 0.001				
Mothers of children with CP								
HADS-A	1.473	1.265	1.715	<0.001				
HADS-D	1.701	1.417	2.042	<0.001	1.439	1.183	1.750	<0.001
BCOS	0.782	0.720	0.850	< .001	0.801	0.734	0.873	<0.001

CP=Cerebral palsy, FMS=Fibromyalgia syndrome, HADS-A=Hospital anxiety scale, HADS-D=Hospital depression scale, VAS=Visual analog scale, FIQ=FMS Impact Questionnaire, BCOS=Bakas Caregiving Outcomes scale, GMFCS=Gross Motor Function Classification System, WeeFIM=Functional independence scale for children

to be higher than that of healthy childcare [3]. Studies have identified the most important predictors of the caregiver burden on parents as the child's degree of disability, parents' anxiety and depression levels, and sense of efficacy [6, 27-29]. Depression and anxiety levels of mothers of children with CP were found to be higher than mothers of healthy children [30, 31]. However, one study found no difference between the anxiety levels of mothers of children with CP and mothers of healthy children [32]. In addition, a positive relationship has been reported between measures of caregiver burden and the severity of anxiety and depression in parents of children with CP [27, 28]. Although there are studies that reached the opposite conclusion [31], other studies reported a direct correlation between the disability and dependency level of the child with CP and the anxiety and stress levels of the mother [33]. Stressful situations associated with

the daily care of a child with CP have been found to, directly and indirectly, affect parents' health, especially mothers [1, 4]. Anxiety and depression are more common in FMS than in the normal population and negatively affect the quality of life [11]. In the current study, although mothers of children with CP needed more caregiver assistance than mothers in the control group, they still had a higher caregiver burden. They had higher levels of depression and anxiety. These results support the results of other studies [3, 30, 31]. Also, in the current study, mothers with FMS of children with CP exhibited higher depression and anxiety scores than those without FMS, and the caregiver burden was higher. The functional level of the children of mothers with FMS was lower, and the dependence level was higher. In addition, there was a direct correlation between the severity of fibromyalgia and the severity of cerebral palsy disease, and the burden of caregiving. The most important factor affecting FMS severity was caregiver burden, followed by maternal depression levels, child's functional status, and the number of siblings with CP. We thought that the heavy burden of providing lifelong care to a child with CP, anxiety, and depression could be triggering factors in the development of FMS by creating physical, environmental, and psychological stressors. Moreover, the fact that children with CP have a high number of siblings have siblings with CP, have low motor functions, and have high dependency levels may have affected the development of FMS by increasing the burden of caregiving.

Limitations

The strength of our study is that it is the first study to investigate FMS in mothers of children with CP. The study's limitations are that it does not reflect the general population, as it is a single-centered cross-sectional study. Nevertheless, we think that it is an essential step in drawing attention to FMS in mothers of children with CP. There is a need for multicenter prospective studies in this area to reflect the general population.

CONCLUSION

This study showed that mothers of children with CP had higher anxiety, depression, and caregiving burden and higher FMS rates than the control group. Children with CP in the group with FMS had a higher number of siblings, a number of siblings with CP, age and dependency levels, and lower motor functions than the children of those without FMS. In addition, this study determined the factors affecting the severity of FMS in mothers of children with CP. These factors are a total number of siblings and number of siblings with CP; decrease in motor functions and increase in dependency levels of children with CP; mothers' anxiety-depression levels; and the increase in caregiver burden. Independent variables are caregiver burden, mothers' depression level, children's GMFCS, and the number of siblings with CP. In the light of these results, we think that Fibromyalgia syndrome can also be kept in mind in the musculoskeletal system pain of mothers of children with CP. Reducing the burden on the mother, who is obliged to take care of her disabled

child for life with psychosocial and physical support, may negatively affect the development of FMS by reducing the stress on mothers. In addition, accurate diagnosis and effective treatment (pharmacological and/or non-pharmacological) can improve mothers' quality of life. Comprehensive studies are needed on this subject.

Authors' Contribution

Study Conception: İAK, MKA, MY; Study Design: İAK, MKA, MY; Supervision: İAK, MKA, MY; Funding: İAK, MKA, MY; Materials: İAK, MKA, MY; Data Collection and/or Processing: İAK, MKA, MY; Statistical Analysis and/or Data Interpretation: İAK, MKA, MY; Literature Review: İAK, MKA, MY; Manuscript Preparation: İAK, MKA, MY and Critical Review: İAK, MKA, MY.

Conflict of interest

The authors disclosed no conflict of interest during the preparation or publication of this manuscript.

Financing

The authors disclosed that they did not receive any grant during conduction or writing of this study.

REFERENCES

- 1. Michael-Asalu A, Taylor G, Campbell H, Lelea LL, Kirby RS. Cerebral palsy: Diagnosis, epidemiology, genetics, and clinical update. Adv Pediatr. 2019;66:189-208. doi: 10.1016/j.yapd.2019.04.002.
- 2. Aisen ML, Kerkovich D, Mast J, et al. Cerebral palsy: clinical care and neurological rehabilitation. Lancet Neurol. 2011;10(9):844-852. doi: 10.1016/S1474-4422(11)70176-4.
- 3. Vadivelan K, Sekar P, Sruthi SS, Gopichandran V. Burden of caregivers of children with cerebral palsy: an intersectional analysis of gender, poverty, stigma, and public policy. BMC Public Health. 2020;20(1):645. doi: 10.1186/s12889-020-08808-0.
- 4. Khayatzadeh MM, Rostami HR, Amirsalari S, Karimloo M. Investigation of quality of life in mothers of children withcerebral palsy in Iran: association with socio-economic status,marital satisfaction and fatigue. Disabil Rehabil. 2013;35(10):803-808. doi: 10.3109/09638288.2012.708818.
- 5. Park EY, Nam SJ. Time burden of caring and depression among parents of individuals with cerebral palsy. Disabil Rehabil. 2019;41(13):1508-1513. doi: 10.1080/09638288.2018.1432705.
- 6. Marrón EM, Redolar-Ripol D, Boixadós M, et al. Burden on caregivers of children with cerebral palsy: predictors and related factors. Univ Psychol. 2013;12(3):767-777. doi: 10.11144/Javeriana.UPSY12-3.bccc

7. Ramezani M, Eghlidi J, Pourghayoomi E, Mohammadi S. Caring-related chronic low back pain and associated factors among mothers of children with cerebral calsy. Rehabil Res Pract. 2020;2020:8854435. doi: 10.1155/2020/8854435.

- 8. Siracusa R, Paola RD, Cuzzocrea S, Impellizzeri D. Fibromyalgia: pathogenesis, mechanisms, diagnosis and treatment options update. Int J Mol Sci. 2021;22(8):3891. doi: 10.3390/ijms22083891.
- 9. Martínez-Lavín M. Fibromyalgia in women: somatisation or stress-evoked, sex-dimorphic neuropathic pain? Clin Exp Rheumatol. 2021;39(2):422-425. doi: 10.55563/clinexprheumatol/0c7d6v.
- 10. Marques AP, Santo ASDE, Berssaneti AA, Matsutani LA, Yuan SLK. Prevalence of fibromyalgia: literature review update. Rev Bras Reumatol Engl Ed. 2017;57(4):356-363. English, Portuguese. doi: 10.1016/j.rbre.2017.01.005.
- 11. Aguglia A, Salvi V, Maina G, Rossetto I, Aguglia E. Fibromyalgia syndrome and depressive symptoms: comorbidity and clinical correlates. J Affect Disord. 2011;128(3):262-266. doi: 10.1016/j.jad.2010.07.004.
- 12. Amital D, Fostick L, Polliack ML, et al. Posttraumatic stress disorder, tenderness, and fibromyalgia syndrome: are they different entities? J Psychosom Res. 2006;61(5):663-669. doi: 10.1016/j.jpsychores.2006.07.003.
- 13. Mutch L, Alberman E, Hagberg B, Kodama K, Perat MV. Cerebral palsy epidemiology: where are we now and where are we going? Dev Med Child Neurol. 1992;34(6):547-551. doi: 10.1111/j.1469-8749.1992.tb11479.x.
- 14. Palisano R, Rosenbaum P, Walter S, Russell D, Wood E, Galuppi B. Development and reliability of a system to classify gross motor function in children with cerebral palsy. Dev Med Child Neurol. 1997;39(4):214-223. doi: 10.1111/j.1469-8749.1997.tb07414.x.
- 15. Ottenbacher KJ, Msall ME, Lyon NR, Duffy LC, Granger CV, Braun S. Interrater agreement and stability of the Functional Independence Measure for Children (WeeFIM): use in children with developmental disabilities. Arch Phys Med Rehabil. 1997;78(12):1309-1315. doi: 10.1016/s0003-9993(97)90302-6. 16. Wolfe F, Clauw DJ, Fitzcharles MA, et al. The American College of Rheumatology preliminary diagnostic criteria for fibromyalgia and measurement of symptom severity. Arthritis Care Res (Hoboken). 2010;62(5):600-610. doi: 10.1002/acr.20140.
- 17. Bennett RM, Bushmakin AG, Cappelleri JC, Zlateva G, Sadosky AB. Minimal clinically important difference in the fibromyalgia impact questionnaire. J Rheumatol. 2009;36(6):1304-1311. doi: 10.3899/jrheum.081090.
- 18. Zigmond AS, Snaith RP. The hospital anxiety and depression scale. Acta Psychiatr Scand. 1983;67(6):361-370. doi: 10.1111/j.1600-0447.1983.tb09716.x.
- 19. Bakas T, Champion V, Perkins SM, Farran CJ, Williams LS. Psychometric testing of the revised 15-item Bakas Caregiving Outcomes Scale. Nurs Res. 2006;55(5):346-355. doi: 10.1097/00006199-200609000-00007.
- 20. Terzi R, Tan G. Musculoskeletal system pain and related fac-

- tors in mothers of children with cerebral palsy. Agri. 2016;28(1):18-24. doi: 10.5505/agri.2015.74436.
- 21. Kaya K, Unsal-Delialioglu S, Ordu-Gokkaya NK, et al. Musculo-skeletal pain, quality of life and depression in mothers of children with cerebral palsy. Disabil Rehabil. 2010;32(20):1666-1672. doi: 10.3109/09638281003649912.
- 22. Czupryna K, Nowotny-Czupryna O, Nowotny J. Back pain in mothers of cerebral palsied children. Ortop Traumatol Rehabil. 2014;16(5):497-505. doi: 10.5604/15093492.1128840.
- 23. Byrne MB, Hurley DA, Daly L, Cunningham CG. Health status of caregivers of children with cerebral palsy. Child Care Health Dev. 2010;36(5):696-702. doi: 10.1111/j.1365-2214.2009.01047.x.
- 24. Rapp CE Jr, Torres MM. The adult with cerebral palsy. Arch Fam Med. 2000;9(5):466-472. doi: 10.1001/archfami.9.5.466.
- 25. Ketelaar M, Volman MJ, Gorter JW, Vermeer A. Stress in parents of children with cerebral palsy: what sources of stress are we talking about? Child Care Health Dev. 2008;34(6):825-829. doi: 10.1111/j.1365-2214.2008.00876.x.
- 26. Farajzadeh A, Maroufizadeh S, Amini M. Factors associated with quality of life among mothers of children with cerebral palsy. Int J Nurs Pract. 2020;26(3):e12811. doi: 10.1111/ijn.12811.
- 27. Gugała B. Caregiver burden versus intensity of anxiety and depression symptoms in parents of children with cerebral palsy as well as factors potentially differentiating the level of burden: a cross-sectional study (Poland). BMJ Open. 2021;11(6):e036494. doi: 10.1136/bmjopen-2019-036494.
- 28. Boztepe H, Çınar S, Ay A, Kerimoğlu Yıldız G, Kılıç C. Predictors of caregiver burden in mothers of children with leukemia and cerebral palsy. J Psychosoc Oncol. 2019;37(1):69-78. doi: 10.1080/07347332.2018.1489441.
- 29. Unsal-Delialioglu S, Kaya K, Ozel S, Gorgulu G. Depression in mothers of children with cerebral palsy and related factors in Turkey: a controlled study. Int J Rehabil Res. 2009;32(3):199-204. doi: 10.1097/MRR.0b013e32832607b6.
- 30. Sajedi F, Alizad V, Malekkhosravi G, Karimlou M, Vameghi R. Depression in mothers of children with cerebral palsy and its relation to severity and type of cerebral palsy. Acta Med Iran. 2010;48(4):250-254.
- 31. Gugała B, Penar-Zadarko B, Pięciak-Kotlarz D, et al. Assessment of anxiety and depression in Polish primary parental caregivers of children with cerebral palsy compared to a control group, as well as identification of selected predictors. Int J Environ Res Public Health. 2019;16(21):4173. doi: 10.3390/ijerph16214173.
- 32. Ones K, Yilmaz E, Cetinkaya B, Caglar N. Assessment of the quality of life of mothers of children with cerebral palsy (primary caregivers). Neurorehabil Neural Repair. 2005;19(3):232-237. doi: 10.1177/1545968305278857.
- 33. Kavlak E, Kara G, Tekin F, Altuğ F, Ök N, Şenol H. Stress and depression levels of mothers who give care to children with cerebral palsy and mental retardation: a comparison study. Pam Med J. 2018;11(1):25-31. doi: 10.5505/ptd.2017.27676