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The Burden of Primary Caregivers of Spinal Muscular Atrophy Patients and Their Needs

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What is already known on this topic?

- *Families of new borns with spinal muscular atrophy (SMA) face several problems and initially report a serious adjustment disorder.*
- *The physical and emotional health of the parents of these children is worse compared to parents of normal and healthy growing children. They need information and psychological and social support.*
- *A family-centered approach has been developed to facilitate the care of children with special needs and to assist their families.*

What this study adds on this topic?

- *Evaluation of family function helps to plan and manage the treatment in line with the family's concerns.*
- *It is important to reveal the problems by evaluating the care burden, needs, and expectations in families of children with SMA, so that this information can be used to plan rehabilitation and interpret the results.*

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ABSTRACT

Aim: This study aims to reveal the problems faced by families of children with spinal muscular atrophy (SMA), by evaluating their care burden, needs, and expectations.

Materials and Methods: The participants were the primary caregivers of 34 children between the ages of 0 and 18 years diagnosed with SMA. Thirteen children were diagnosed with type 1, 13 children with type 2 and 8 children with type 3 SMA. Data on the medical history, functional levels of the participants, and the characteristics of families were collected. The childrens' parents completed the Family Needs Survey and the Zarit Caregiver Burden Scale.

Results: According to the results of the Family Needs Survey, it was found that information was the most common requirement, and this was independent of the level of education. According to the Caregiver Burden Scale, it was recorded that 64.7% of the caregivers were under mild/moderate burden. While there was a moderate correlation ($r = 0.574$; $P < .001$) between the Caregiver Burden Scale and the Family Needs Survey, it was observed that the functional level of the child was not associated with family needs and caregiver burden.

Conclusions: Our study suggests that the needs of families of SMA patients, especially related to income level, have changed. The caregivers' burden is not directly related to the income level or the functional level of the child. Families' need for information should also be prioritized within the rehabilitation program.

Keywords: Care burden, family needs, social support, spinal muscular atrophy

INTRODUCTION

Spinal muscular atrophy (SMA) is a group of neuromuscular diseases involving the anterior horn cells of the spinal cord and the motor nuclei of the brainstem. It progresses with programmed cell death and leads to symmetrical weakness/atrophy of voluntary muscles in the whole body, leading to loss of strength, regression in mobility, and impairment of pulmonary functions due to the involvement of respiratory muscles.[1-3] It is divided into 4 subtypes according to the age of onset and the severity of muscle weakness. SMA type 1 (Werdnig–Hoffmann disease), which is the earliest and most severe form, is diagnosed with severe respiratory problems and hypotonia within the first 6 months of life. Usually, children die before 2 years of age due to progressive respiratory failure. SMA type 2 (subacute form) starts between 6 and 18 months, even though the symptoms are similar to type 1, they do not worsen quickly. SMA type 3 (Kugelberg–Welander disease) is also known as childhood SMA and onset is after 2 years of age. Usually, the first symptom is difficulty walking, and it is the mildest. Generally, there is no shortening in the patients' lifespan. Type 4 begins in adulthood.[2,4,5]

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Families confronted with these problems that occur with the birth of the baby or shortly after birth primarily experience a serious adjustment disorder.[6] Patients and their families are heavily stressed when the course of the disease and the absence of a definitive cure are announced. Physical and emotional health of the SMA patients' parents are worse than those of the parents of normal and healthy growing children. They need information, and psychological and social support.[7]

The role of the family is very important in the lives of children with disabilities. Family-oriented care has been developed to facilitate the care of children with special needs and to assist their families. The main features of the family-centered approach are the ideas that children are best known by their families, the families are specialized in their children's needs, and each family is unique; family and community support is provided so that the child's functionality is at an appropriate level.

Evaluation of family function helps to plan and manage the treatment in line with the family's concerns. The education of the family, the sociocultural structure, and the psychological approach of the parents play an important role in the development of the child with SMA.

This study aims to investigate the care burden, needs, and expectations of families with children diagnosed with SMA. The study hypothesizes that the problems can be revealed by evaluating the care burden, needs, and expectations of families. Thus, it is possible to benefit from this information in planning rehabilitation and interpretating the results.

METHODS

Children between the ages of 0 and 18 diagnosed with SMA, who applied to the Marmara University Pendik Training and Research Hospital, Department of Physical Medicine and Rehabilitation, Pediatric Rehabilitation Special Polyclinic Polyclinic between July 2019 and December 2019, were included in the study. Approval was obtained from the Marmara University Pendik Education and Research Hospital Ethics Committee (09.2019.728) and registered on ClinicalTrials.gov with NCT04228718 number.

In the power analysis performed with G * Power 3.1.9.2 (means: difference from constant – one sample case) to determine the number of the sample, the effective size was calculated based on a previous study[8] on children with SMA. Based on the Zarit Caregiver Burden Scale, 0.53 was taken, and the sample size of the study was calculated as 30 with 80% power and 0.05 α error.

To reveal the caregiver burden of SMA patients on their primary caregivers, the Zarit Caregiver Burden Scale (CBS: ZBI), consisting of 22 items, which was developed by Zarit et al.[9] with the validity study conducted by Özlü et al.,[10] was applied. With this scale, which can be used to evaluate the distress experienced by caregivers of individuals in need of care, the caregiver/patient relationship, the health status of the caregiver, psychological comfort, social life, and economic burden are evaluated. A minimum score of 0 and a maximum score of 88 can be obtained on the scale. The higher the score, the higher the distress experienced. İnci et al.[11] adapted the scale to Turkish and tested its validity and reliability.

To determine the requirements, the "Family Needs Assessment Tool" (FNAT:FNS) was used, which was developed by Bailey and Simeonsson (1988), adapted into Turkish by Sucuoğlu (1995) and re-tested for validity and reliability by Cavkaytar et al.[13] (2014). In the study by Sucuoğlu,[12] the scale consisted of 35 items. After the validity and reliability study by Cavkaytar et al.[13] (2014), the number of items in the FNAT was reduced to 29. FNAT is a 3-point Likert scale instrument. FNAT is rated "1" (absolutely no), "2" (not sure), and "3" (absolutely yes). FNAT does not contain reversed score items, so by directly adding the scores given to the items, the number of family needs can be determined over the total score. The lowest score that can be obtained from FNAT is 29 and the highest score is 87. It can be said that as the scores obtained from the scale and subscales increase, the families' levels of need also increase.

Motor function for SMA type 1 in clinical studies is evaluated with The Children's Hospital of Philadelphia Infant Test of Neuromuscular Disorders (CHOP-INTEND), which provides a sensitive measurement for significant changes.[14,15] CHOP-INTEND uses a 0-64 point scale. Higher scores indicate better motor function. It has been used to reliably measure the natural motor function decline in babies with SMA type 1.[16,17]

The Hammersmith Functional Motor Scale (HMFS) expanded is a scale developed by O'Hagen et al.,[18] used to evaluate motor functions in SMA type 2 and 3. It contains 33 items scored 0, 1, and 2, with a total of 66 points.

An "Evaluation Form" containing the medical history, accompanying pathologies, and family information of the patients was filled in for each patient. CBS and FNAT questionnaires were applied to the families after the evaluation form was filled out. The children were then taken to a quiet room where they could feel comfortable, to minimize distraction, and were evaluated with functionality tests.

Statistical Analysis

Statistical analysis of the study was performed using the SPSS (IBM Statistical Packages for the Social Sciences; Armonk, NY, USA) Version 20.0. The information is presented in the tables (Table 1-4). The value for statistical significance was accepted as $P < .05$. Numerical data were expressed as mean ± standard deviation (mean ± SD), median and lowest-highest, while categorical data were expressed as number (n) and percentage (%).

Differences between groups were analyzed using the Mann–Whitney U test when they did not provide parametric properties. Tests between SMA types were compared with the Kruskal–Wallis test. Analysis of variance: fixed effects, omnibus, one way, was used to calculate the post hoc power ($1 - b$ error probability) for the 2 evaluations used in the comparison between types. The Spearman correlation coefficient was used for inter-test correlations and was evaluated as <0.30 = can be rejected; 0.30-0.50 = low; 0.50-0.70 = medium; 0.70-0.90 = high; >0.90 = very high.[19,20]

Table 1. Demographic Characteristics of Families	
Variable	***n* (%)**
Mother's level of education	
Primary school	13 (38.2)
Secondary school	8 (23.5)
High school	9 (26.5)
University	3 (8.8)
Postgraduate	1 (2.9)
Father's level of education	
Primary school	6 (17.6)
Secondary school	6 (17.6)
High school	13 (38.2)
University	7 (20.6)
Postgraduate	2 (5.9)
Level of ıncome (TL)	
<3000	17 (50)
3000-5999	12 (35.3)
6000-9999	4 (11.8)
>15 000	1 (2.9)
Number of siblings	
None	7 (20.6)
1	14 (41.2)
2	7 (20.6)
3	5 (14.7)
4	1 (2.9)
Another disabled child in the family	
Yes	9 (26.5)
No	25 (73.5)

RESULTS

Thirteen patients were diagnosed as type 1, 13 were type 2, and 8 were type 3 SMA mean age was 7.61 ± 6.23; 25 (73.5%) were male and 9 (26.5%) were female. The questionnaires were filled out by mothers for 22 children, fathers for 11 children and the grandmother for 1 child. It was determined that only 14.7% of the mothers were working. The mean age of mothers was 35.11 (20-50) years, and the mean age of fathers was 39.36 (25-58) years. The parents were all married. It was determined that in 97.1% of the cases, the father provided a living, and the monthly income of half of the families was between 0 and 2999 Turkish lira (Table 1).

According to the FNAT results, it was observed that the caregivers primarily needed information (Table 2). Under this heading the need for information about the institutions the child could benefit from in the future (97.1%) was prominent. It was found that the need for information was not related to the educational level of the mother or father ($P = .305$; $P = .429$). It was observed that as the total income increased, the need for information relatively decreased ($r = -0.431$; $P = .011$).

Under the heading of general support and social service, the need for someone to talk to about problems in the family (50%) was at the forefront.

Under the heading of environmental disclosure, the family's need to learn how to support each other in difficult times (47.1%) was at the forefront.

Under the financial requirement heading, the need to provide special tools for the child (70.6%) and to meet the costs of therapy, special education, care, etc. (70.6%) were prominent. It was observed that as the total income increased, the financial need decreased ($r = -0.550$; $P = .001$).

According to the CBS results, 22 people were registered under light/medium load, 9 people under medium/high load, 2 people under no/little load, and 1 person under overload.

The CBS total scores were moderately correlated with the FNAT total score. Looking at the relationship between CBS and FNAT subcategories, there was a moderate correlation with the need for information score, a low level of correlation with general support and social service score, a low level of correlation with environmental disclosure, and there was no correlation with financial need (Table 3).

Considering the relationship of the scales with SMA types, there was no significant difference between the types ($P > .05$) (Table 4). In type 1, as age and number of siblings increased, the need for information decreased ($r = -0.600$, $P = .030$; $r = -0.581$, $P = .037$); as the need for general support and social service increased, the need for environmental disclosure increased ($r = -0.832$, $P < .001$). The environmental disclosure was found to be more difficult for boys ($P = .036$). The need for information about the institutions (92.3%) that the child can benefit from in the future was prominent.

In type 2, as the age increased, the total score of CBS decreased ($r = -0.557$, $P = .048$); as the total income increased, the information and financial needs decreased ($r = -0.632$, $P = .020$; $r = -0.624$, $P = .023$). It was observed that there were more financial needs for girls ($P = .043$). Moreover, the presence of another disabled child affected the FNAT total score ($P = .009$) and subcategories except financial needs ($P = .087$) ($P < .05$). There was a high correlation between the CBS total score and the FNAT total score and the need for information ($r = -0.763$, $P = .002$; $r = -0.763$, $P = .002$), and a moderate correlation with other subcategories ($P < .05$). The need for information (100%) about the institutions that the child could benefit from in the future was the prominent item.

In type 3, as the number of siblings increased, material needs increased ($r = 0.709$, $P = .049$). A high correlation was found between the CBS total score and the FNAT total score, the need for information, and the need for general support and social service ($r = -0.843$, $P = .009$; $r = -0.843$, $P = .009$; $r = 755$, $P = .031$). The prominent needs were the those regarding the child's situation, about the institutions that the child could benefit from now and in the future, the need to read materials such as books and articles written about parents who have similar children, and the need for financial assistance to meet the costs of the child's therapy, special education, care, etc. (100%).

When we look at the relationship between the scales of the CHOP applied in type 1 and the Hammersmith Functional Motor Scale : HMFS scores applied for type 2 and 3, there was no correlation between the functionality levels of FNAT and CBS.

Table 2. Family Needs Assessment Tool	
Items of Need	**Yes (%)**
1. I need more information about the child's situation.	73.5
2. I need information about how I can control my child's behavior.	61.8
3. I need information on how to teach some skills to my child.	64.7
4. I need information on how to play/talk with my child.	50
5. I need information about the institutions my child can benefit from now.	82.4
6. I need information about institutions my child can benefit from in the future.	97.1
7. I need information about how children grow and develop.	74.7
8. I need to read materials such as books and articles written about parents who have children with similar characteristics as my child.	73.5
9. I need more time to devote to myself.	47.1
10. I need help finding a dentist who can help my child.	47.1
Need for ınformation	**66.2**
11. I need someone in my family to talk to about my problems.	50
12. I need help finding a caregiver who can take care of my child when necessary.	29.4
13. I Need help finding a preschool or kindergarten for my child.	41.2
14. When I attend a meeting, I need help finding a nursing home or kindergarten where my child can receive appropriate care.	32.4
15. My family needs help to make decisions about who will do housework, childcare, and other work.	32.4
16. My family needs help to make decisions about recreational activities and things to do.	35.3
General support and social service	**36.8**
17. I need to talk to religious officials to find solutions to my problems.	38.2
18. I need help explaining my child's situation to siblings.	32.4
19. I need help in explaining my child's situation to my spouse and my wife's family,	17.6
20. My spouse needs help to understand my child's situation.	23.5
21. When my neighbor, friend, or a stranger asks about my child's situation, I need help with how to respond.	26.5
22. I need help explaining my child's situation to other children.	17.6
23. I need help in learning how our family can support each other in difficult times.	47.1
Environmental disclosure	**29**
24. I need assistance to cover my expenses such as food, house rent, medical assistance, clothing, transportation.	44.1
25. I need help to provide special tools for my child.	70.6
26. My child needs help to cover therapy, special education, care, etc.	70.6
27.My wife and I need help to find a job.	23.5
28.Sometimes I need help to pay the caregiver who cares for my child.	32.4
29. I need help to get the toys that my child needs.	35.3
Financial need	**46.1**

DISCUSSION

The term caregiver burden generally includes not only the physical burden related to care but also the psychological, social, and financial burden experienced while providing care.[21,22] Children with SMA have significant physical disabilities in daily life. This situation causes parents to experience emotional symptoms such as needing time for themselves, having more care burden, restricting other family members, feeling sad for the child, and exhaustion.[23,24] Based on these reasons, our study evaluated the care burden and needs of families.

The majority of those who completed the questionnaire were women, and mothers in particular, which indicates that women assume more responsibilities in the care of disabled individuals, similar to data from the literature.[8,25-29] The economic status of the families and the educational levels of the parents were also compatible with previous studies conducted with families with disabilities.[30-32]

There is no study in the literature which details the FNAT as it was used in our study, in patients with SMA. However, there were studies on disabled people reported previously.[13,33-35] When the FNAT results were evaluated in our study, the total mean score

Table 3. Correlations					
	Need for Information	**General Support and Social Service**	**Environmental Disclosure**	**Financial Need**	**FNAT Total**
CBS	$r = 0.579$, $P < .001$	$r = 0.490$, $P = .003$	$r = 0.492$, $P = .003$	$r = -0.262$, $P = .135$	$r = 0.574$, $P < .001$
*With the Spearman correlation test. CBS, Caregiver Burden Scale; FNAT, Family Needs Assessment Tool.					

Table 4. Comparison of Scales Between Types and Their Subscales

	SMA Type	*n*	Median (Percentile) (25-75%)	Mean ± SD	*df*	χ^2	*P*	Significant Difference
CBS total	Type 1	13	33 (26-41)	33.84 ± 9.13	2	1.913	.384	No difference
	Type 2	13	32 (24.5-37)	30.76 ± 12.20				
	Type 3	8	38.5 (28-57)	41.25 ± 14.88				
Need for information	Type 1	13	24 (22-27)	24.30 ± 3.70	2	1.951	.377	No difference
	Type 2	13	24 (17-30)	23.46 ± 6.17				
	Type 3	8	28 (24.5-29.75)	26.75 ± 3.73				
General support and social service	Type 1	13	11 (6-12)	10.46 ± 3.79	2	2.182	.336	No difference
	Type 2	13	12(7.5-14)	11.30 ± 3.94				
	Type 3	8	15 (9.25-16.75)	13.25 ± 4.33				
Environmental disclosure	Type 1	13	10 (9-13)	11.15 ± 3.60	2	4.993	.082	No difference
	Type 2	13	11 (8-13)	11 ± 3				
	Type 3	8	15 (11-18.75)	14.87 ± 4.05				
Financial need	Type 1	13	12 (9-16)	12.23 ± 3.85	2	4.127	.127	No difference
	Type 2	13	11 (8.5-14)	11.38 ± 3.50				
	Type 3	8	14.5 (12.5-17.5)	14.62 ± 2.77				
FNAT total	Type 1	13	56 (50-64)	58.15 ± 10.27	2	4.831	.089	No difference
	Type 2	13	62 (42-67.5)	57.15 ± 14.66				
	Type 3	8	71.5 (58.75-80.75)	69.5 ± 12.38				

*Kruskal–Wallis test.
df, degrees of freedom; χ^2, chi-square; SD, standard deviation; CBS, Caregiver Burden Scale; FNAT, Family Needs Assessment Tool.

was 60.44 ± 13.24 (33-85). It was also observed that as the total income increased, the total scores of FNAT and all its subcategories decreased. Öztürk reported a statistically significant difference only between financial need and economic status.[36]

In the evaluation between subcategories and demographic information, it was seen that the need for information was not related to the educational level of the parents. It may be explained by the lack of knowledge about the disease, which is not related to the level of education. The other possible explanation is that every individual concerned is anxious to learn about the illness, and this is common regardless of their level of education.

In our study, the gender of the child, the gender of the caregiver, the educational level, the number of siblings, and the presence of other disabled children did not affect the needs. However, with regard to the type of SMA, it was more difficult for boys to make environmental disclosure in type 1, and families with girls in type 2 stated more financial needs. When we look at the literature, there is one study that has results similar to ours,[36] while the other stated that mothers with male children need more financial resources.[37] Akçatepe and Kargın also stated in their study on hearing-impaired children that the needs do not vary according to educational level and age.[37,38]

Ersoy Quadır et al.[34] found the total average of FNAT above 60 points in their study and stated that mothers with low educational levels felt the need for the family the most. It has been observed that among mothers, those with a low level of education, a low family income, and a more children have higher financial needs. The results of our study were similar to these results, however, it was determined that only the total income level affects the need in families of children with SMA. Again in the same study,[34] it has been reported that mothers with low educational levels need information and have difficulty disclosing their child's disability to the environment. In our study, the educational level was found to be only related to the need for information and total income level. In both our study and the reference study, there was no relationship between demographic evaluations and general support and social service need. The reason why the results of all FNAT differ from the previous study may be due to the different sample sizes and the populations studied.

When the subcategories and items of the FNAT were examined, it was observed that families of children with SMA mostly needed information. This result was similar to the literature.[35-37] According to the results of FNAT, in the study by Karadağ-Saygi et al.,[35] the need for information in families was observed the most. Again under this heading, the need for information about the child's situation and the institutions that can benefit the child currently and in the future come to the fore.

Similarly, Kaytez et al.[33] reported that more than half of the families answered "yes" in all of the sub-items regarding need for information sub-items. These results are in line with our study.

Again in the same study, "yes" rates are reported below 50% for all items in the sub-items of general support and social service and environmental disclosure.[33] In another similar study, the same result was obtained in all sub-items except general

support (56.8%).[35] In our study, one of the items of general support and social service, the item "need someone who can talk about problems in the family" was the primary item, with half of the families saying "yes", and all other results were similar to the previous study.

In the sub-items of financial need, the need to provide the special tools required for the child and to meet the costs of therapy, special education, care, etc. for the child were prominent. In a similar study in the literature, these items were those with a "yes" answer above 50%.[33]

When we look at the evaluation among the subcategories of FNAT, the scale total score showed a high correlation with all subcategories. While the categories of knowledge, environmental disclosure, and general support social service needs show a medium-high correlation between each other, there was only a low correlation between financial need and all other subcategories. This indicates that the parents' needs for information related to the disease, environmental disclosure, and social support are the same regardless of the financial situation.

When we look at the results between types in SMA, there was no significant difference. It can be said that this result is due to the difference in diagnosis and age of onset of the diseases.

Considering that the socioeconomic burden is more heavy than the psychological burden of the disease, several tools have focused on caregiver burden in SMA.[8,23,39,39,40]

The CBS was used in our study to assess the caregiving burden. CBS total mean score was 34.41 ± 12.17 (9-63). There are 2 studies in the literature regarding the use of this scale in SMA patients.[8,40] This result is similar to results of a previous study.[8]

When the CBS results were categorized, 1 person was recorded under overload, 9 people under medium/severe load, 22 people under light/medium load, and 2 people under no/little load. Although these results show the burden of care for children with SMA, it can be said that these families have started to see their children as a normal part of their lives, as the results of low and light-medium burden are predominant in the evaluation. A study in the literature stated that, although the care given to these children is a stress factor, parents can see their children as a necessary and normal part of their lives, to ensure their well-being.[39]

When the demographic characteristics of the caregivers were compared with the CBS, no correlation was found with the child's age, caregiver's age, educational status, number of siblings, and income status. Dambi et al. did not find a significant correlation between the age and educational status of the caregiver and the care burden in their study of children with cerebral palsy, similar to our study.[41-43] The differences in the subject may have been caused by the differences in the populations included in the study.

When we look at the literature on the relationship of CBS with educational status, Coşkun et al.[42] reported that similar to our results, educational status does not affect the burden of care. In the literature, studies suggest that educational status increases or decreases the care burden.[44,45]

Artan et al. reported in their study that as the number of children increased, the caregivers' burden increased.[46] Again, the reason for these different results of our study may be that the families who participated in our study mostly had 1 or 2 children other than the child with SMA.

Similar to our study, a significant statistical difference was not found between income levels and caregiver burden in another study.[46] On the contrary, in 2 other studies, it was reported that the low income level is one of the factors that increase caregiver burden.[47,48] This may be because most of the families participating in our study are families with low income levels.

When the CBS average score was compared with the gender of the child and the gender of the caregiver, there was no significant difference in terms of gender in both cases. It is similar to data from literature that children do not affect the care burden in terms of gender. Although in our study, the gender-related burden of care did not change in caregivers, studies in the literature report a higher care burden on mothers more than fathers.[41,43,46] This may be due to the unequal number between the genders included in our study.

Considering the correlation of the tests with each other, a moderate correlation was observed between CBS and FNAT results, while CBS was not correlated with financial needs. We observed a low correlation with general support, public service, and environmental disclosure, and a moderate correlation with the need for information. This result confirmed that the burden on the caregiver increased with increasing needs. However, it showed that this burden was not caused by financial needs and that it was a caregiver burden entirely due to the need for information related to the disease, in addition to environmental disclosure and social support situations related to the disease. A similar study also reported that as the needs of families increased, their burden increased.[36]

In a study conducted by Piran et al. among children with chronic diseases with various levels of disability, they looked at the correlation between the age and educational status of the caregiver and the child's age with CBS and found no correlation, similar to our study. However, they found a negligible relationship between CBS and total income ($r = -0.140$, $P = .027$).[21]

Similarly, a study also reported that families of children with SMA experienced high levels of stress and strain, with decreased social support, the severity of the disease, and behavioral problems.[23]

The literature suggests that effective tools such as everyday activities and caregiver burden should be combined and a functionally evaluated to provide additional information about gains and losses.[49]

In our study of a certain sample, we evaluated the types with functionality scales within the group, and we did not find any correlation between FNAT and CBS. This may be due to the small sample size.

In our inter-type post hoc power analysis, the power value was found in the medium (power: 0.46-0.54) power range according to the CBS and FNAT total scores. This suggests that our

study has a modest probability of accurately detecting a true effect. This can be shown as the limitation of our study.
In conclusion, our study showed that the increased needs of families of SMA patients, especially concerning their income level, changed, but the caregiver burden was not related to income level. It was observed that the needs of the children with SMA in the family, especially due to disease-related reasons, affect the caregiver burden.

Ethical Committee Approval: Ethical committee approval was received from the Ethics Committee of Marmara University, (Approval No: 09.2019.728).

Informed Consent: Written informed consent was obtained from all participants who participated in this study.

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