

## BURDEN OF CAREGIVERS OF PATIENTS WITH DUCHENNE MUSCULAR DYSTROPHY: RELATIONSHIP TO FUNCTIONAL CAPACITY

Sobrecarga de cuidadores de pacientes com Distrofia Muscular de Duchenne: relação com a capacidade funcional

Sobrecarga de los cuidadores de pacientes con Distrofia Muscular de Duchenne: relación con la capacidad funcional

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### How to cite this article:

Eduardo IM, Oliveira LC, Ribeiro MFM, Prudente COM. Burden of caregivers of patients with Duchenne Muscular Dystrophy: relationship to functional capacity. 2021 jan/dez; 13:547-552. DOI: <http://dx.doi.org/10.9789/2175-5361.rpcf.v13.9302>.

### ABSTRACT

**Objective:** To relate a functional capacity of patients with Duchenne muscular dystrophy to the burden of their family caregivers. **Methods:** A cross-sectional study with 35 patients and their family caregivers from a rehabilitation center in Goiânia-GO. Were used for the functional evaluation of the Vignos Scale, Motor Function Measure Scale, Barthel Index, Egen Klassification Scale and for burden evaluation the Burden Interview. **Results:** The caregivers had moderate burden, with a mean of 23,6 ( $\pm$  10,3). Regarding the patients, the average score of the instruments showed that they were in an advanced stage of disease staging and impairment of functional capacity. No significant relationship was found between the caregiver burden and the patient's functional capacity. **Conclusion:** The caregiver is directly connected to the care function of the patient, so the multiprofessional team needs to pay attention to their mental health, offering support for specialized interventions.

**Descriptors:** Neuromuscular diseases; Duchenne muscular dystrophy; Quality of life; Motor activity; Caregivers.

### RESUMO

**Objetivo:** Relacionar a capacidade funcional de pacientes com Distrofia Muscular de Duchenne com a sobrecarga dos seus cuidadores familiares. **Métodos:** Estudo transversal com amostra composta por 35 pacientes e seus cuidadores familiares, de um centro de reabilitação de Goiânia-GO. Foram utilizados para avaliação da capacidade funcional a Escala de Vignos, Medida da Função Motora, Índice de Barthel e Escala *Egen Klassification* e para avaliação da sobrecarga o *Burden Interview*. **Resultados:** Os cuidadores estavam

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com sobrecarga moderada, com média de 23,6 ( $\pm 10,3$ ). A pontuação média dos instrumentos demonstrou que os pacientes estavam em fase avançada de estadiamento da doença e prejuízo da capacidade funcional. Não foi encontrada relação significativa entre a sobrecarga dos cuidadores e a capacidade funcional dos pacientes. **Conclusão:** O cuidador está diretamente ligado à função de zelar do paciente, portanto a equipe multiprofissional precisa se atentar também à saúde mental deste, oferecendo suporte para intervenções especializadas.

**Descritores:** Doenças neuromusculares; Distrofia muscular de Duchenne; Qualidade de vida; Atividade motora; Cuidadores.

## RESUMEN

**Objetivo:** Relacionar la capacidad funcional de pacientes con Distrofia Muscular de Duchenne con la sobrecarga de sus cuidadores familiares.

**Métodos:** Estudio transversal con muestra compuesta por 35 pacientes y sus cuidadores familiares, de un centro de rehabilitación de Goiânia-GO. Se utilizaron para la evaluación de la capacidad funcional la Escala de Vignos, Medida de la Función Motora, Índice de Barthel y Escala Egen Klassifikation y para evaluación de la sobrecarga el Burden Interview.

**Resultados:** La sobrecarga de los cuidadores fue considerada moderada, con promedio de 23,6 ( $\pm 10,3$ ). En cuanto a los pacientes, la puntuación media de los instrumentos demostró que estaban en fase avanzada de estadificación de la enfermedad y perjuicio de la capacidad funcional. No se encontró una relación significativa entre la sobrecarga de los cuidadores y la capacidad funcional de los pacientes. **Conclusión:** El cuidador está directamente ligado a la función de velar del paciente, por lo tanto el equipo multiprofesional necesita atenderse también a la salud mental de éste, ofreciendo soporte para intervenciones especializadas.

**Descriptores:** Enfermedades neuromusculares; Distrofia muscular de Duchenne; Calidad de vida; Actividad motora; Cuidadores.

## INTRODUCTION

The most common muscle diseases in childhood are dystrophinopathies, among which Duchenne Muscular Dystrophy (DMD) stands out.<sup>1</sup> DMD is considered the most frequent, most severe and the most rapidly evolving form. It is a recessive genetic pathology linked to the X chromosome, which causes a mutation in dystrophin, a protein that prevents the degeneration of normal muscle fiber, promotes its maintenance and guarantees an effective muscle contraction.<sup>2</sup>

DMD is typically diagnosed in Brazil between seven and 11 years of age. Children are born healthy and evolve in development, but close to five years of age, muscle weakness is evident. The progression of the disease varies from individual to individual, functional losses include symptoms such as delayed gait development, accumulation of adipose tissue, loss of lean mass and progression to wheelchair addiction.<sup>3</sup> Overall, most individuals survive until the age of 20 and die due to heart or respiratory failure.<sup>4</sup>

The multidisciplinary health team needs to closely analyze the clinical evolution of DMD, monitoring the effectiveness of treatments and considering the possibility of changing behaviors according to the patient's needs.

This analysis of clinical evolution is made possible by the application of functional scales.<sup>5</sup> Functional capacity is an important aspect that works as an indicator of the subject's degree of independence, for carrying out activities of daily living. Preventive measures and therapeutic interventions are essential to reduce the individual's disabilities.<sup>6</sup>

The physical and functional impairments that patients have also interfere in the health of their caregivers, as the family suffers and needs to learn to deal with the feelings aroused by the disease.<sup>4</sup> Caring for a person with DMD is associated with overload, as shown in the literature.<sup>7-9</sup> Throughout the disease's progression, the fragility of the patient's health and mental state also brings suffering, anxiety and depression to the caregiver.<sup>7</sup>

The literature makes clear the need for a holistic approach aimed at the patient's family and the development of new studies, aiming at expanding knowledge for the benefit of family caregivers' health. These surveys should serve as an incentive for the development of support actions to promote the quality of life of these individuals and reduce the burden.<sup>7-9</sup>

There is a shortage of pragmatic guidelines regarding the care that must be taken both in the care of patients and in caregivers. There is a need for the professionals' commitment to address this caregiver's mental health, especially when the patient's physical health and mental state are impaired. Therefore, the objective of the current study was to relate the functional capacity of patients with DMD to the burden of their family caregivers.

## METHOD

Cross-sectional study, with a quantitative approach, developed in a reference center for rehabilitation in the city of Goiânia-GO. The convenience sample consisted of 35 patients who were undergoing treatment, with a confirmed diagnosis of DMD and their informal family caregivers. Patients with other associated neurological diseases and non-family caregivers were excluded.

For the evaluation of functional capacity, the Vignos scale, the Motor Function Measure (MFM), the Egen Klassifikation scale (EK) and the Modified Barthel Index were used; and to assess the burden of caregivers, the Burden Interview. In addition, caregivers answered a sociodemographic and clinical profile card with personal information about themselves and patients, developed by the researchers.

The Vignos scale ranks the function from 0 to 10, and the higher the score, the worse the functional performance. It is a simple and easy to apply instrument. Functional activities are aimed at lower limbs, which are the most affected with the evolution of the disease.<sup>10</sup>

The MFM evaluates the function in a static and dynamic way, composed of 32 items, divided into three dimensions: dimension 1 (D1): standing position and transfers; dimension

2 (D2): axial and proximal motor function; dimension 3 (D3): distal motor function. Each item is scored from 0 to 3, zero being unable to start the task, one just starting, two performing partially or imperfectly and three performing the task completely. The total and partial scores for the dimensions are expressed in percentages in relation to the maximum score, which ranges from 0 to 100.<sup>11</sup>

The Modified Barthel Index is an instrument that assesses the subject's level of independence to perform ten basic life activities. Each item is assessed individually, ranging from 1 to 5. The total score is classified as follows: 10-total dependency; 11-30 severe dependence; 31-45 moderate dependency; 46-49 slight dependence and 50 total independence.<sup>12</sup>

The EK scale is divided into 10 categories, each with four items (0 to 3). The total sum of the items varies from zero to 30, with zero characterizing minimal impairment and 30 greater degree of impairment in peripheral muscle strength, contractures and respiratory function.<sup>13</sup>

The *Burden Interview* is a 22-item instrument that assesses the burden of caregivers related to the functional and behavioral disability of the beneficiary of care. Each item on the scale is scored from 0 to 4, with 0 = never, 1 = rarely, 2 = sometimes, 3 = very often, 4 = almost always. The total score is obtained by adding all the items and can vary from 0 to 88. The higher the total score, the greater the overload.<sup>14</sup> The determination of the overload classification score is done as follows: <21 = little or no overload, 21-40 = mild to moderate overload, 41-60 = moderate to severe overload, 61> severe overload.<sup>15</sup> Caregivers answered the sociodemographic and clinical profile form, the Modified Barthel Index and the Burden Interview. The patients were submitted to physical evaluations, in order to complete the Vignos scale, MFM and EK scale.

The data were analyzed using a significance level of 5% ( $p < 0.05$ ). The characterization of the profile of patients and caregivers was done through absolute (n) and relative (%) frequency and descriptive statistics considering the continuous variables. The parametry of the data was verified using the Shapiro-Wilk normality test. The comparison of the Burden Interview with the profile of patients and caregivers was performed using Student's t-tests and analysis of variance (ANOVA). Pearson's Correlation was used to verify the relationship between the Burden Interview and the MFM, the Modified Barthel Index, the EK scale and the Vignos scale.

The research followed the Regulatory Guidelines and Norms for Research involving human beings (Resolution 466/12, of the National Health Council) and was approved on May 17, 2016, by the Research Ethics Committee of PUC Goiás, opinion No. 1,547 .418. Data collection took place from July to December 2016, at the institution itself. First, the patients and their caregivers were selected according to the criteria and all information regarding the research was clarified. The application of the procedures started after the process of consent and signing of the terms of consent and consent by the participants.

## RESULTS AND DISCUSSION

DMD patients had a mean of 13.8 ( $\pm 3.9$ ) years of age. The symptoms started on average at 4.0 ( $\pm 2.1$ ) years and the loss of gait at 9.5 ( $\pm 2.3$ ) years of age. Of the total sample, 31 (88.6%) did not walk and 27 (77.1%) attended school regularly.

Table 1 presents the results of the assessment of functional capacity, using the instruments. The patients had an advanced stage of disease staging, assessed by the Vignos Scale; and severe dependence, according to the Modified Barthel Index score. As for the evaluation of MFM, they indicated better performance of distal motor function (D3) and worse in standing motor function and transfers (D1). Peripheral muscle weakness and respiratory function, assessed by the EK scale, showed a level of impairment slightly below the instrument's average.

**Table 1** - Result of functional capacity assessments. Goiânia, GO, Brazil, 2016

	Median	Average $\pm$ SD	Minimum	Maximum
Vignos Scale	7,0	7,1 $\pm$ 1,4	3,0	9,0
MFM				
D1	0,0	10,2 $\pm$ 25,3	0,0	100,0
D2	58,3	60,6 $\pm$ 28,3	2,8	100,0
D3	81,0	73,2 $\pm$ 23,3	4,8	100,0
MFM total	40,6	42,6 $\pm$ 22,4	2,1	100,0
EK Scale	13,0	13,3 $\pm$ 5,9	0,0	27,0
Barthel Index	28,0	28,7 $\pm$ 9,1	13,0	49,0

Caregivers were on average 38.7 ( $\pm 8.2$ ) years old and had an average monthly income of R\$ 1985.7 ( $\pm 2262.2$ ) reais. Table 2 describes the sociodemographic profile of caregivers. It is noted that the majority are female, married or living maritally, do not work, receive social benefits, have the car as their main means of transportation, have leisure activities and do not perform psychological counseling. Most of them have an elementary school education level.

**Table 2** - Description of the sociodemographic profile of the caregivers. Goiânia, GO, Brazil, 2016

Caretaker profile	N	%
<b>Sex</b>		
Female	32	91,4
Male	3	8,6
<b>Marital status</b>		
Married	15	42,9
Single / divorced	8	22,9
Lives maritally	12	34,3

Caretaker profile	N	%
<b>Education</b>		
Elementary School	17	48,6
High school	15	42,9
University education	3	8,6
<b>Employment</b>		
Formal	5	14,3
Informal	5	14,3
Not working	25	71,4
<b>Social Benefits</b>		
No	7	20,0
Yes	28	80,0
<b>Transportation</b>		
Car	20	57,1
Car and bus	1	2,9
Moto	1	2,9
Bus	13	37,1
<b>Leisure Activity</b>		
No	15	42,9
Yes	20	57,1
<b>Psychological treatment</b>		
No	28	80,0
Yes	7	20,0

Regarding the burden of caregivers, assessed by the Burden Interview, the average score was 23.6 ( $\pm 10,3$ ), which represents mild to moderate burden. As for the classification, the majority presented mild to moderate overload (Table 3).

**Table 3** - Classification of caregiver burden. Goiânia, GO, Brazil, 2016

Overload (Burden Interview)	N	%
Little or none	14	40,0
Light to moderate	19	54,2
Moderate to severe	2	5,7

Table 4 correlates the total Burden Interview score with the other instruments for assessing functional capacity. It is observed that there was no statistical significance between the instruments.

**Table 4** - Correlation between caregiver burden and patients' functional capacity. Goiânia, GO, Brazil, 2016

	Burden Interview	
	R	p
MFM1	0,17	0,32
MFM2	0,10	0,56
MFM3	-0,08	0,65
MFM total	0,14	0,43
Barthel Index	0,01	0,95
EK Scale	0,01	0,95
Vignos Scale	-0,11	0,53

*p* = Pearson's correlation; *r* = Correlation coefficient

The present study presented a sample of caregivers composed mainly of female individuals, with low education and who do not have an employment relationship. The literature points out that these are characteristics common to caregivers who have an overload.<sup>16</sup> In this study, the sample was classified with moderate overload, according to the evaluation by the Burden Interview. Burden analysis is important to assess not only caregivers, but also the needs of care beneficiaries.<sup>16</sup>

Only three studies were found that used methodological bases similar to the current one to investigate the burden. These researchers found an average of 26.38, 297 and 28.315 points in the Burden Interview, which represents mild to moderate overload and converges with the result of the present study, of 23.6 points.

The literature demonstrates that the presence of overload in caregivers of children with DMD is associated with reduced quality of life<sup>8</sup> and with predictors of stress such as anxiety and distress. Several factors contribute to this relationship, such as an increase in household expenses, few leisure hours and the requirement of the care process, associated with other daily activities.<sup>7</sup>

In a study carried out in Mexico, with a sample composed of 74% of caregivers of individuals with DMD, the researchers concluded that parents of children with neuromuscular diseases have depressive symptoms of indecision, pessimism, guilt and suicidal ideation. The authors also noted that there was no association between these symptoms and the patient's clinical characteristics.<sup>18</sup>

Despite the burden, caregivers of patients with DMD value their activity and report feeling of satisfaction, in addition

to facing this situation as a mission to be accomplished.<sup>4</sup> This positive attitude in this scenario is often related to trust in religion and family emotional support they receive, and these are factors that reduce overload.<sup>9</sup>

As for patients, the sample was composed mainly of individuals in advanced stage of disease staging and impaired functional capacity. In the investigated literature, only one research was found with the sample profile similar to the current one, which related functional dependence through the score obtained in the MFM, with the burden of caregivers, through the Burden Interview.<sup>8</sup>

As in the current study, the authors found no association between the variables, functional capacity and overload. However, they concluded that caregivers for older children were more overwhelmed. That is, the burden was related to the time of care and not to less functional independence.<sup>8</sup>

Although it was not the objective of this study, there are other clinical characteristics of the patient that can influence the burden of these caregivers and which are described in the literature: presence of tracheostomy, need for mechanical ventilation<sup>17</sup> and older children.<sup>9</sup> It is noteworthy that patients in the the present study did not present severe impairment of respiratory function, assessed by the EK scale, none had a tracheostomy and most were in wheelchairs.

As for the burden assessment instrument, although this is the most used questionnaire in the current literature to assess caregivers of patients with dystrophies, it is important to note that there is a need for a validated instrument for this population. The questions and alternative answers that are present in the instrument may not take into account the impact of the subjective burden of each caregiver<sup>7</sup>, that is, they may not be suitable for this sample.

It is important to note that therapeutic interventions demonstrate attenuation of overload and associated feelings.<sup>16</sup> The support of social networks and support professionals are essential, as they help to reduce stress factors, through coping strategies.<sup>19,20</sup> In the current study, only 20% of caregivers performed psychological counseling, and 42.7% did not have leisure activities. Caregivers of individuals with chronic illnesses need emotional support, as they need someone to listen to them, be empathetic, provide comfort, security, confidence and dedication.<sup>21</sup>

## CONCLUSION

Individuals with DMD showed impaired functional capacity and their caregivers were classified as having moderate overload. However, overload is not related to aspects of patients' functional capacity. There is little literature that investigates the association between these variables, therefore, future research is necessary to improve the professional approach aimed at both the patient and the caregiver.

This study contributes to the scientific environment, as it points to a knowledge gap that is little investigated and broadens the view on the provision of informal care.

The family member is directly linked to the integral and intensive care of the patient, so the multidisciplinary team also needs to pay attention to the patient's mental health, offering support and referral for evaluation and specialized interventions, in order to reduce the burden. It is worth highlighting the need for an instrument that enables a better understanding of the needs of caregivers of patients with muscular dystrophies.

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Received in: 02/09/2019

Required revisions: 24/10/2019

Approved in: 04/02/2020

Published in: 20/04/2021

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have no conflict of interest.**