

Quality of Life of Family Caregivers of Patients with Duchenne Muscular Dystrophy

Qualidade de Vida de Familiares Cuidadores de Pacientes com Distrofia Muscular de Duchenne

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Abstract

Duchenne Muscular Dystrophy (DMD) is the most common type of dystrophy. The care provided to the patient with DMD is exhausting and can affect the quality of life (QoL) of caregivers. The objective of the study was to describe the QoL of family caregivers of patients with DMD, relating to sociodemographic and clinical aspects of the patients. This is a cross-sectional quantitative study performed at a rehabilitation institution in Goiânia, Goiás. The sample consisted of 30 family caregivers and their patients with DMD. The instruments applied were: sociodemographic questionnaire; Scale Vignos; and Quality of Life Measure of the World Health Organization (WHOQOLbref). The mean age of the caregivers was 39.20 and the patients were 14.07 years. Most of the caregivers were female, had primary or secondary education. Symptoms of the disease began on average at 3.87 years of age. The mean score in the Vignos Scale was 7.13. The psychological domain of the WHOQOLbref had the highest mean (66.11%) and the lowest environment (55.52%). There was a negative correlation between the age of onset of symptoms and the psychological domain ($r = -0.44$; $p = 0.01$). There was a significant relationship between the transport medium and the environment domain ($p = 0.05$). Family caregivers present moderate QOL. The earlier the patients initiate the more favorable symptoms is the psychological aspect of the caregiver's QOL, suggesting that the earlier the diagnosis is the better the caregiver's QoL.

Keywords: Quality of Life. Caregivers. Muscular Dystrophy, Duchenne.

Resumo

A Distrofia Muscular de Duchenne (DMD) é o tipo de distrofia mais comum. O cuidado prestado ao paciente com DMD é desgastante e pode afetar a qualidade de vida (QV) dos cuidadores. O estudo teve como objetivo descrever a QV de cuidadores familiares de pacientes com DMD, relacionando com aspectos sociodemográficos e clínicos dos pacientes. Trata-se de um estudo quantitativo, transversal, realizado em uma instituição de reabilitação de Goiânia - Goiás. A amostra foi composta por 30 cuidadores familiares e seus pacientes com DMD. Os instrumentos aplicados foram: questionário sociodemográfico; Escala Vignos; e Medida de Qualidade de Vida da Organização Mundial de Saúde (WHOQOL- bref). A média de idade dos cuidadores foi de 39,20 e dos pacientes de 14,07 anos. A maioria dos cuidadores era do sexo feminino, possuía ensino fundamental ou médio. Os sintomas da doença iniciaram em média com 3,87 anos de idade. A média da pontuação na Escala Vignos foi de 7,13. O domínio psicológico do WHOQOL- bref apresentou maior média (66,11%) e o meio ambiente a menor (55,52%). Houve correlação negativa entre a idade de início dos sintomas e o domínio psicológico ($r = -0,44$; $p = 0,01$). Houve relação significativa entre o meio de transporte e o domínio meio-ambiente ($p = 0,05$). Os cuidadores familiares apresentam moderada QV. Quanto mais cedo os pacientes iniciam os sintomas mais favorável é o aspecto psicológico da QV do cuidador, sugerindo que quando mais precoce é o diagnóstico melhor é a QV do cuidador.

Palavras-chave: Qualidade de Vida. Cuidadores. Distrofia Muscular de Duchenne.

1 Introduction

Duchenne Muscular Dystrophy (DMD) is one of the most devastating genetically neuromuscular diseases¹. It is the most common type of dystrophy affecting 1:3800 to 1: 6200 boys born alive². Clinical manifestations usually begin in childhood³, muscle weakness is progressive and irreversible, evolving to inability to ambulate between nine and 13 years of age⁴. After gait loss, patients are confined to a wheelchair, aggravating the deformities of the spine and lower limbs, interfering with functional independence and self-esteem⁵.

In the last decades, improvement in treatment conditions has enabled these patients to have a higher life expectancy⁶.

Still, there is no cure, which makes functional dependence more dependant according to the evolution of the disease, influencing the quality of life of patients and their caregivers^{1,3}. DMD causes disability, regardless of the clinical aspects related to it, affecting the emotions of family caregivers, family life and social life⁷. The worst functional capacity of patients worsens the quality of life of caregivers⁸. According to the World Health Organization (WHO), quality of life consists of physical, mental, social well-being and not only in the absence of diseases or illnesses⁹.

The appearance of the disease in a family, leading the sick member to dependence, leads to the need to modify their daily

life activities, and the need for a caregiver, usually the family is the main care provider¹⁰. The care given to an individual with chronic disease is highly stressful and can affect the physical and psychological well-being of caregivers. As fundamental role is played in the patient's life, the caregiver needs to maintain his or her well-being and a healthy life, receiving health care and guidance to provide care to the needy family. In this sense, the assessment of the quality of life of the caregiver and the factors that may influence his or her reduction becomes relevant for the health care professional to know his or her needs, draw goals and schedule actions to minimize the negative aspects of the care process¹¹.

Most studies are aimed at the quality of life of patients with DMD, and publications with family caregivers are restricted. An intervention should be considered based on the whole family and not just on the child with DMD. In addition, when children are diagnosed with DMD, parents should be instructed to share the responsibility of caring for the child in order to improve their quality of life and reduce the burden.

The objective of this study was to analyze the sociodemographic profile and quality of life of family caregivers of patients with DMD treated at a rehabilitation institution in Goiânia and to relate sociodemographic aspects of caregivers and clinicians of patients with the quality of life of family caregivers.

2 Material and Methods

It is a descriptive, quantitative and cross-sectional research. Data were collected at a reference center in rehabilitation in Goiânia. The sample consisted of 30 patients and their family caregivers. Inclusion criteria were patients with confirmed diagnosis of DMD and their family caregivers; and those who were at the time of data collection in rehabilitation process at the institution. Patients with other associated neurological diseases and professional caregivers were excluded.

The instruments applied to caregivers were: A sociodemographic profile sheet containing general information about patients and their caregivers; Vignos Scale to evaluate the stage of patient staging; And the quality Of Life Measure of the World Health Organization (*WHOQOL-bref*) to evaluate the quality of life of the caregiver. Analysis of medical records was performed to collect information on the characteristics of each patient.

WHOQOL-bref is an instrument used to assess the quality of life of adult populations. It contains 26 questions, of which 24 are distributed in four domains: physical, psychological, social relations and the environment. In addition, the instrument presents two general questions, one referring to the perception of quality of life and the other to satisfaction with health. The results of the gross scores of each facet were transformed into a score ranging from zero to 100. The closer the mean score is to 100, the more positive the perception of the overall quality of life is¹².

The patients were submitted to physical evaluations to fill the Vignos Scale. The scale classifies the function from 0 to 10, and the higher the rating, the worse the functional performance. This is a simple and easy method to apply. The functional activities are aimed at lower limbs, which are the most affected by the evolution of the disease¹³.

The data were analyzed using *the Statistical Package of Social Sciences (SPSS, 23.0)*. In all statistical analyzes, a significance level of 5% was adopted ($p \leq 0.05$). The sociodemographic and clinical profile description of the group was performed by means of absolute and relative frequency for categorical variables and descriptive statistics for continuous variables. The Shapiro-Wilk test was applied to verify whether the continuous variables presented a normal distribution. Not being verified the normality of data non-parametric tests were applied. Spearman's correlation was used to verify *the relationship of WHOQOL-bref domains* with the interviewer's age, with the Vignos Scale, monthly income, and the onset of symptoms. The comparative analysis of *WHOQOL-bref domains* with sex, schooling and means of transport was performed using the non-parametric Mann-Whitney and/or Kruskal-Wallis test.

This research was approved by the Research Ethics Committee of the Pontifical Catholic University of Goiás (PUC Goiás), according to opinion 1.547.418. It was carried out in accordance with the recommendations of Resolution 466/12 of the National Health Council. All participants over the age of 18 read and manifested the acceptance by signing the free and informed consent form, and for patients under the age of 18 the consent term before data collection.

3 Results and Discussion

The sample consisted of 30 family caregivers, with mean age of 39.20 years ($SD \pm 8.32$). The average monthly family income was 2095.33 ($SD \pm 2424.20$) reais. The majority of caregivers are female (93.30%), have high or primary education (90.00%), and have no work activity (70.00%). There was a higher percentage of people who claim to be practicing religion (76.7%); having their own residence (53.3%); and receiving some kind of social benefit (80.0%). Most have some kind of leisure activity (63.3%) and car is a means of transport (63.3%). Only 23.3% of caregivers have psychological treatment and 16.7% have chronic diseases. In 83.3% of the cases, the caregiver reported receiving support from the partner in the care of the DMD patient.

The mean age of patients with DMD was 14.07 years ($SD \pm 4.08$) and 73.3% were enrolled in some type of school. Caregivers reported that the age of manifestation of DMD symptoms varied from 1.0 to 7.0 years, with the mean age of 3.87 ($SD \pm 1.93$) years. These patients were unable to walk on average with 9.52 ($SD \pm 2.36$) years of age, and three patients were still unable to walk. On the Vignos scale, the mean score of the patients was 7.13 ($SD \pm 1.41$), with a minimum of 3.0 and a maximum of 9.0 points.

Table 1 shows the description of WHOQOL-bref of DMD patients caregivers. The physical, psychological and social domains showed approximate averages. The worst-average domain was the environment.

Table 1 - Description of the domains and general score of the caregivers' WHOQOL-bref, n=35, Goiania, GO, Brazil, 2018

WHOQOL-bref	Mean	Standard Deviation	Minimum	Maximum
Physical domain	65.60	12.21	42.9	92.9
Psychological domain	66.11	16.91	29.2	91.7
Social domain	65.83	20.57	25.0	91.7
Environment domain	55.52	18.55	25.0	96.9
Overall quality of life	63.27	13.37	32.6	82.5

Source: Research data.

Table 2 shows the correlation between caregiver's age, Vignos scale, monthly income and symptom onset age with WHOQOL-bref domains and total score. It was observed that there was a negative correlation between the age at which the patients' symptoms started with DMD and the psychological domain of WHOQOL-bref.

Table 2 - Correlation among caregiver's age, Vignos scale, monthly income and symptom onset age with WHOQOL-bref domains and total score

WHOQOL-bref	Caregiver's age	Vignos scale	Monthly Income	Age at onset of symptoms
Physical domain	r = 0.01 p = 0.97	r = 0.02 p = 0.90	r = 0.03 p = 0.87	r = -0.21 p = 0.27
Psychological domain	r = 0.34 p = 0.06	r = 0.17 p = 0.36	r = 0.16 p = 0.40	r = -0.44 p = 0.01*
Social domain	r = 0.20 p = 0.29	r = 0.10 p = 0.59	r = 0.23 p = 0.21	r = -0.07 p = 0.72
Environment domain	r = 0.05 p = 0.81	r = 0.11 p = 0.55	r = 0.31 p = 0.10	r = -0.01 p = 0.96
Overall quality of life	r = 0.26 p = 0.17	r = 0.20 p = 0.29	r = 0.23 p = 0.22	r = -0.18 p = 0.34

*Spearman Correlation

Source: Research data.

Table 3 shows the variables gender, education and transportation with the domains and total WHOQOL-bref score. There was a significant relationship between the transport means and the environment domain. Care givers who use csar have a better quality of life in the environment domain.

Table 3 - Relationship among sex, education and means of transport with WHOQOL-bref domains and general score, n=35, Goiânia, GO, Brazil, 2018.

Variables	Physical	Psychological	Social	Environment	General QV
Sex*	p = 0.27	p = 0.80	p = 0.19	p = 0.93	p = 0.56
Female	65.05 ± 12.23	66.22 ± 16.37	64.58 ± 20.62	55.47 ± 19.05	62.83 ± 13.57
Male	73.21 ± 12.63	64.58 ± 32.41	83.33 ± 11.79	56.25 ± 13.26	69.35 ± 11.63
Schooling**	p = 0.57	p = 0.11	p = 0.66	p = 0.46	p = 0.34
Elementary School	63.74 ± 9.65	60.26 ± 16.72	64.74 ± 19.88	50.72 ± 17.22	59.86 ± 11.75
High School	66.33 ± 15.07	68.45 ± 17.04	68.45 ± 21.97	58.26 ± 20.05	65.37 ± 15.74
Higher Education.	70.24 ± 8.25	80.56 ± 2.41	58.33 ± 22.05	63.54 ± 17.21	68.17 ± 3.47
Means of transport*	p = 0.51	p = 0.87	p = 0.91	p = 0.05	p = 0.66
Car	65.79 ± 11.12	68.42 ± 11.22	67.54 ± 19.82	61.51 ± 16.96	65.82 ± 9.53
Bus	67.50 ± 13.09	64.58 ± 23.75	66.67 ± 19.64	47.19 ± 16.63	61.48 ± 16.50

*Mann-Whitney; **Kruskal-Wallis

Source: Research data.

Studies show that mothers are responsible for the childcare^{14,15}, similar to that found in this study. Women are usually historically and culturally related to the domestic/maternal sphere while man is responsible for formal work. In contradiction to the reality found in Brazil, a study carried out in India with caregivers of children with DMD showed that parents are the main caregivers, and this is due to a cultural factor, where the woman is responsible for taking on the formal role of work and the man the family commitments¹⁷.

In the present study, most caregivers did not develop formal work activity. Researchers assessed stress on caregivers of patients with DMD and showed that work outside the home is a predictor of high stress, suffering and dissatisfaction with life¹⁵.

The caregivers, in their majority, were married and

reported that they received support from the companion in the care of the DMD patient. In another study carried out in Italy that evaluated the family context of patients with DMD, it showed that the difficulties and the caregiver burden are higher when the latter is single¹⁸. According to another study carried out with mothers of children with DMD, the married women had less stress when compared to the single ones. This data suggests that social support provided by a partner plays a fundamental role in the well-being of the caregiver¹⁹.

In this study, a small part of the caregivers received psychological treatment. Another study stated that prolonged care can primarily result in feelings of sadness, loss and concern about the future of the patient. These feelings tend to develop depression and other minor psychiatric disorders²⁰.

In the present study, the patients presented mean age at

onset of symptoms around three to four years. This age group is typically described in the literature²¹. The mean age that patients were unable to walk was 9.52 years. This information is in accordance with a study carried out in France that brought an average age for the loss of walking around nine years and five months. Another study carried out in the United Kingdom showed higher values, around 13 years of age.

A study that used the Vignos Scale to quantify the patients' functional independence showed that those unable to ambulate have an average score from 7 to 8 points on the scale²⁴. This information is in accordance with the present study, which analyzed in its majority dependant patients and consequently with a higher mean (7.13) in the Vignos Scale.

Regarding the quality of life of caregivers, they presented a higher average in the psychological domain (66.11%) and lower in the environment (55.52%). The closer the mean score is to 100, the more positive the perception of the perception of quality of life is¹². Few studies have investigated the burden of caregivers of children with DMD. A Brazilian study observed that poor quality of life is explained by the overload of caring for children with DMD. Still in this study, almost 40% of caregivers reported being dissatisfied with their lives⁸. In addition, stress, depression and anxiety have been described in groups of caregivers of children with DMD¹⁵.

The psychological domain showed a significant correlation with the age of onset of symptoms. The sooner the patients present the symptoms of the disease, the better the psychological aspect of the quality of life of the family caregiver. This suggests that caregivers present good adaptation strategies when the disease in the family comes earlier, knowledge about the disease and consequently early diagnosis can enable caregivers to receive adequate support and guidance. Adaptation of an individual to an event is a process based on primary and secondary cognitive assessment²⁵. The primary assessment refers to the understanding of what the disease is, while the secondary one implies the development of emotional strategies to deal with the difficulties of caring. Therefore, adaptation can be influenced by internal factors, such as the attitude of the family caregiver toward the patient, and external resources, such as the availability of social and professional support²⁶. A qualitative study carried out in Italy addressed the main positive aspects related to the care of patients with DMD and showed that caregivers of patients with longer disease time recognized more positive aspects of the care process with greater conviction²⁷.

The environment domain encompasses physical security, financial resources, leisure and transport. An expressive part of the caregivers reported having some kind of leisure activity. A study that outlined the consequences of prolonged care in patients with DMD, presented that more frequently cited difficulties were to stop having hobbies and lack of leisure activities.

In the present study, there was no correlation between sociodemographic aspects and the quality of life, but carers

have a better quality of life in the environment domain. A study carried out in South Korea analyzed the quality of life of caregivers on the influence of sociodemographic aspects and showed that the family charge is significantly influenced by the sociodemographic characteristics of the relatives, by the clinical severity of the patient, by financial resources, by the level of education, social and emotional aspects²⁰.

There were no significant correlations between the patient's functional dependence and the caregiver's quality of life. A study carried out in Sao Paulo that used the same instrument to evaluate the quality of life of the caregiver showed that the burden is mainly influenced by the patients' age. Caregivers of older patients had more burden than those of younger patients, suggesting that the care time of these patients overloads the caregivers and consequently decreases their quality of life. It also clarifies that the origin of the burden is specifically related to the time of care of people and is not correlated with the loss of functional independence of patients⁸.

4 Conclusion

Family caregivers of patients with DMD present a compromise in the quality of life, especially in relation to the environment domain. The sooner the patients present the symptoms of the disease, the better the psychological aspect of the quality of life of the family caregiver. Health professionals should recognize the quality of life as a problem of interference in the relationships of family caregivers with patients with DMD. An intervention based on the whole family and not just on the child with DMD should be considered. This study presents as a source of encouragement for the development of new research, studies with a larger sample could provide us with additional information on these caregivers, which would enable the development of family intervention strategies.

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