

Experiences of caregivers of boys with Duchenne Muscular Dystrophy: “When I look at him, my world falls apart”

Experiencias de cuidadoras de niños con Distrofia Muscular de Duchenne: “Cuando lo volteo a mirar, se me une el cielo con la tierra”

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Resumen

La Distrofia Muscular de Duchenne (DMD) es una enfermedad neuromuscular, clasificada como una enfermedad huérfana o rara, transmitida por la madre, y que conlleva al deterioro físico progresivo y fatal de quienes la desarrollan, que en su mayoría son hombres. Aun con los tratamientos farmacológicos disponibles, el pronóstico de vida de los pacientes sigue siendo desalentador. Los niños van perdiendo progresivamente la movilidad y funcionalidad hasta que fallecen. En consecuencia, el progreso de esta enfermedad requiere de cuidados frecuentes o permanentes que implican altos niveles de exigencia para las cuidadoras de niños y de jóvenes que han desarrollado la DMD. El apoyo proveniente del sistema de salud es

escaso para ambos, lo que se traduce en una intensa sobrecarga para las madres y mujeres de la familia que son quienes, habitualmente, cuidan a la persona diagnosticada con DMD durante su vida.

Este estudio cualitativo de tipo fenomenológico tuvo como objetivos analizar el significado de la experiencia de ser cuidadora de niños y jóvenes que viven con DMD en Colombia, develando el sentido que tiene para las mujeres familiares asumir el rol de cuidadora. Se entrevistaron siete mujeres entre los 25 y los 64 años encargadas de cuidar, por lo menos, a un niño o joven con diagnóstico de DMD, previo consentimiento informado. Adicionalmente, los cuidadores desarrollaron un ejercicio de asociaciones libres.

Para el análisis de datos se realizó un análisis temático de las narrativas, encon-

trando tres temas fundamentales alrededor de los cuales se le da sentido a esta experiencia: (1) descubrir la enfermedad, (2) vivir en función del paciente y (3) reconstruir el sentido de vida. Así, en un momento inicial las cuidadoras se enfrentan al diagnóstico de la enfermedad brindado por los médicos, quienes les explican su curso y su naturaleza. La cuidadora inicia un proceso de duelo, en el que la tristeza, la negación y la resignación tienen lugar, y emociones como la frustración y la impotencia se presentan en su máxima expresión. La manera en la que es comunicado el diagnóstico es evaluada por las cuidadoras como poco sensible. De hecho, ninguna de las participantes en el estudio recibió apoyo psicológico durante el proceso.

En segundo lugar, ser la figura de cuidado implicó cambios a nivel personal, familiar y social, para lograr responder a las necesidades de los niños con DMD. En este sentido, algunas debieron renunciar a sus empleos, alejarse de amistades e incluso algunas manifestaron que se han separado de su pareja o han vivido experiencias de deterioro en las relaciones con su pareja y familia extensa. Además, junto con sus hijos han vivenciado experiencias de discriminación, vulneración de derechos y escaso apoyo social, experiencias que son factores de riesgo para la salud mental.

Finalmente, la enfermedad termina por constituirse en el elemento vinculante entre ellas y sus niños. Los hallazgos de la presente investigación permiten identificar que, en el proceso de enfermedad, se produce una reconstrucción del sentido de vida. Así, las estrategias de afrontamiento se dirigen a lograr una transformación espiritual. La presencia de la enfermedad es interpretada como una manera de ser probadas y desarrollar sus cualidades como mujer y como madre. Entonces, el cuidado y la relación madre-hijo, son componentes motivacionales que transforman y dan sentido a la vida.

En conclusión, los resultados muestran que el significado de la DMD está fuertemente

vinculado a un proceso de intenso sufrimiento y estrés psicológico, con alta carga negativa.

En particular, durante los primeros años después del diagnóstico, que debe ser asimilado y elaborado para aprender a convivir con la DMD y la certeza de la muerte prematura de un hijo.

Palabras clave: cuidadores, mujeres, distrofia muscular de Duchenne, investigación cualitativa, análisis fenomenológico interpretativo, análisis temático, niños con discapacidad.

Abstract

Duchenne muscular dystrophy (DMD) is a neuromuscular disease that leads to progressive and fatal physical deterioration. The progression of this disease requires frequent or permanent care that implies high levels of demand for the caregivers of children who have developed DMD. Certainly, this disease affects diagnosed children and their caregivers as well. The support from the health system is scarce for both, which translates into an intense burden for the mothers and women of the family who care the person diagnosed with DMD during their lifetime. This qualitative study with phenomenological approach aimed to analyze the meaning of the experience of being a caregiver for boys and young men living with DMD in Colombia. After obtaining their informed consent, caregivers were interviewed and a thematic analysis of their narratives was performed to determine three key topics that endow their experiences with meaning: (1) finding out about the disease, (2) living according to the needs of boys and young men, and (3) providing a new meaning to life. The results show that the meaning of DMD is strongly associated with intense suffering and psychological stress, with a high negative burden, especially during the initial years after diagnosis. The study suggests that it is important to assimilate and process these experiences and findings to learn how to live with DMD and identify the certainty of a child's premature death.

Keywords: caregivers, women, Duchenne muscular dystrophy, qualitative research, interpretative phenomenological analysis, thematic analysis, disabled children.

Introduction

Duchenne muscular dystrophy (DMD) is a neuromuscular (Barros, Moreira, & Rios, 2018) and genetic (D'Amario et al., 2017) pathology associated with the X chromosome; therefore, men develop DMD, whereas women are generally its carriers (Han et al., 2020). DMD has been classified as an orphan or rare disease (Barros, Moreira, & Ríos, 2018; Crisafulli et al., 2020), with a pooled global prevalence of 7.1 (95 % CI: 5.0 - 10.1) and birth prevalence of 19.8 (95 % CI: 16.6–23.6) per 100 000 males (Crisafulli et al., 2020). Typically, it is diagnosed in early childhood (Birnkranz et al., 2018), and its progression is severe, subsequently evolving into cardiorespiratory failure in early adulthood (Łoboda & Dulak, 2020). Currently, DMD is not curable (Messina & Vita, 2018), but kids with this diagnosis can exceed the second decade of their lives (Birnkranz et al., 2018). The available treatment is palliative care until symptomatic control is achieved through interdisciplinary and multidisciplinary management (Thomas et al., 2014; Messina & Vita, 2018; Muñoz-Rivas et al., 2020).

DMD compromises the functionality of boys (Guiraud et al., 2015) through a chronic and degenerative course (Landfeldt, Edström, Buccella, Kirchner, & Lochmüller, 2018), which is characterized by generalized muscle weakness, loss of strength and balance, problems in motor skill development, and movement restriction (Türk et al., 2018). Children with DMD stop walking (Birnkranz et al., 2018) approximately between the ages of 10 and 12. In some children, their cognitive ability is also compromised (Anikiej, Mánski, & Bidzan, 2018; Naidoo & Anthony, 2020), while in others it is preserved intact (Rana et al., 2016).

The course of DMD demands frequent and specialized care, which is highly time-consuming for the caregivers of boys and young men with DMD (Gocheva et al., 2019). Women usually feel identified with caregiving and accept it as an act of love or a natural and moral duty; they assume that caregiving is their responsibility (Kleinman, 2012). They even perceive caregiving as a positive act (Magliano et al., 2014); in regards to this, caregivers can build resilience by continuously coping with difficulties (Glover, Hendron, Taylor, & Long, 2018).

Although caregivers play an essential role in the care of boys and young men with DMD, the review of literature shows the consequences in their mental health. For example, this dynamic of care exceed the resilience of caregivers (such as having children with comorbidities or several disabled children), and training of caregivers—usually parents, specifically mothers or women (Arias & Muñoz-Quezada, 2019)—can trigger conditions that diminish their quality of life (Lue, Chen, & Lu, 2018). These conditions include caregiver's burnout (Gérain & Zech, 2018), difficulties related to the time and energy needed to provide the best care for their children (Thomas et al., 2014), mood alterations such as depression (Landfeldt et al., 2018) and anxiety (Landfeldt et al., 2016), deterioration of their social interaction and self-care (García-Calvente, 2004), interpersonal and family functioning problems, difficulties in daily care (Thomas et al., 2014), sleep disorders that lead to physical and mental health disorders (Nozoe et al., 2017), pain, discomfort and difficulties in performing their daily activities (Landfeldt et al., 2016), high levels of distress and stress (Kenneson & Bobo, 2010) and negative impact on the mental health (Lue et al., 2018).

Furthermore, in their daily experience the relationship with the health system is difficult, given that there are different levels of trust and information involved and a struggle to achieve the children's need that are not in

line with the need to receive compassionate and competent treatment (Dawson & Kristjanson, 2003). Unlike the health system in countries such as India, the health system in Colombia covers the costs of treating children with DMD, as long as the child is formally linked to the system and receives timely care. However, similar to India, the economics of scarcity is a feature shared by several families (Thomas et al., 2014) in Colombia that affects their quality of life and limits timely access and consultation of their health problems. Although the literature shows that research on caregivers of kids with DMD has increased, it is still scarce, especially in Latin American countries.

Additionally, Aguilar-Delgado and Ramos-Lira (2020) in a literature review study of articles published between 2008 and 2018 with a psychosocial perspective on DMD, identified a predominant use of quantitative approach and three themes around which studies with caregivers of children with DMD have been developed: The impact on the quality of life of mothers who assume the role of primary caregivers; the experience of female carriers; and the preparation of parents for their children's transition from adolescence into adulthood. Although these are guiding findings, they do not provide an understanding of the caregiving experience. For this reason, a phenomenological approach would contribute toward a search for the meaning of the experiences and thus provide a foundation from which to build an understanding of what it is like to be a female and mother caregiver for boys or young men who live with DMD.

Also, the literature on the meaning of the experiences of being a caregiver for children and young people living with DMD is limited, and those available findings apply to other socio-cultural contexts (ej. Samson et al., 2009; Yamaguchi, Sonoda, & Suzuki, 2019). However, these studies have demonstrated the value of knowing what women and mothers feel and experience in the process of being caregivers for family members with DMD.

For example, the qualitative study by Samson et al. (2009) has found that parents of children with DMD experience a transformation in the way they interpret the disease. Initially, they perceive it as a stranger and later, as part of their child, going through depressive states, accepting this reality and finally, everyone adapting to it.

Phenomenology can thus result in valuable knowledge about individuals' experiences. From this approach, we can analyze questions about: What is the experience of being a caregiver of children living with DMD like? What does it mean for female caregivers to take on this role? Within this framework, in this study we set out to answer the question of: What are the meanings of the experience of being a caregiver in Colombia for boys and young men with Duchenne Muscular Dystrophy?

Method

Design

The present research is a qualitative research of phenomenological approach. The focus in this study is the experiences, senses, and meanings (Bautista, 2011). This study addressed the experiences of being a caregiver of boys and young men with DMD living in Bogotá, Colombia.

Study participants

The participants were seven women aged between 25 and 64 who provided care to one or two living children; only two participants were in an existing common law marriage, and the remaining participants, except one participant, were separated after diagnosis of the disease in their children. The boys and young men, who were taken care of by female caregivers, were between the ages of 9 and 17, only one of them was 21 years old. The caregivers of boys and young men attending genetic consultation at a private healthcare provider. All the seven caregivers provided informed

consent, which was assessed previously and approved by the Research Ethics Committee. Therefore, the caregivers participated in this study on a voluntary basis, and their anonymity was preserved.

Instruments

The techniques used to obtain the informa-

tion were individual unstructured interview and free associations incorporated into the interview process (Table 1). The duration of the interview was variable between 40 and 90 minutes for each caregiver at an office. We performed as many interviews as required to ensure data saturation. The narratives were recorded in audio and then transcribed.

Table 1

Stimulus-based questions that comprised the unstructured interview.

Introduction

At the beginning of the interview, we aimed to strengthen the participation of caregivers, highlight the benefit of their participation, and motivate them to narrate their experiences, by showing them the researchers' obvious interests. In this context, the instructions were as follows:

As we have told you, this study is very important for us because we aim to generate more chances to support patients with DMD and their caregivers. We appreciate your participation in the study. We ensure that we will provide all our time required to willingly listen to everything you want to share with us about your experience while dealing with this disease. Everything you tell us is valuable and relevant, and we believe it is our responsibility to listen to you without any judgments.

Stimulus Questions

(The questions were asked only if narrative of the caregivers did not cover these topics)

Tell us your experience with (patient's name) who has this disease.

How did you find out about the disease?

How has life been since you learned about this condition of (patient's name)? Please explain this in detail.

What words come to your mind when I say DMD?

What has been the most difficult thing during this process?

According to you, what are your main concerns about the disease?

Have you ever wondered why this happened? And what are your thoughts about this?

Would you say that there has been a pre and post diagnosis of (patient's name) disease? Or has this disease of (patient's name) changed your life? And why?

Who helps you in the caregiving of (patient's name)?

10. Describe your relationship with (patient's name).

What is (patient's name) like?

What is the best thing about your relationship?

What is the most difficult concern in your relationship?

11. Describe the relationship of (patient's name) with the doctors

12. Describe your relationship with the doctors

13. Who do you speak with and share your feelings about the disease of (patient's name)?

Data analysis

In line with the phenomenological approach, a thematic analysis was carried out (Braun & Clark, 2012). Data analysis was conducted using the Nvivo software, version 12. In the process, the data was segmented into codes on the basis of their meanings and then regrouped into the topics that best described the essence of the experience (Flores-Kanter & Medrano, 2019; Willig, 2008). In addition, free associations were also analyzed, by grouping the associated words or phrases into categories. Also, based on their responses, we created a word cloud using the Nvivo 12.

To develop the thematic analysis, we followed the phases proposed by Braun and Clark (2012): (1) Familiarization with the information: each narrative was carefully studied through several readings in which we began assigning initial codes for a visualization of meanings and patterns, looking for a complete approach to the content. (2) Generation of initial codes: the fragments of the transcripts were coded and segmented into topic codes using the Nvivo software, version 12.

(3) Searching for topics: codes were grouped into topics and subtopics. (4) Reviewing the topics: topics were reviewed and refined by reading all the extracts contained in them to determine their coherence within a pattern. Furthermore, we reviewed the thematic map and verified whether it reflected the meanings found in the data set.

Results

In order to promote the understanding of the analysis and the interaction of the different transitions of the experiences and meanings of the caregivers, we built an interpretative scheme (Figure 1). In this scheme the topics identified were repeatedly grouped at each moment in the narratives of the caregivers, which reflect the perspectives they have elaborated on their experience. The results were grouped into three topics that represent different moments of the caregiving process: (1) Finding out about the disease, (2) Living according to the children's needs, and (3) Providing a new meaning to life.

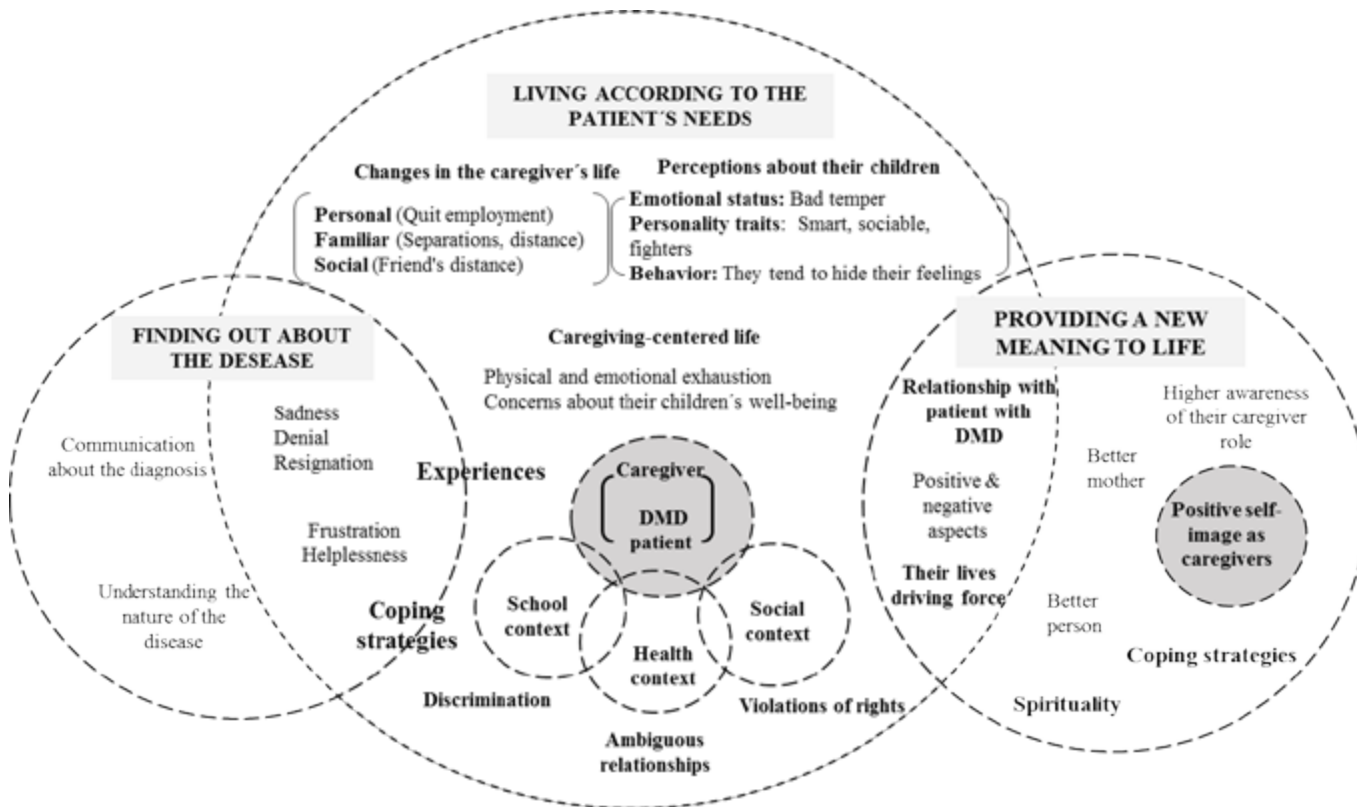


Figure 1. Comprehensive scheme of thematic analysis.

Finding out about the disease

Initially, the family members are informed about the diagnosis of the disease by doctors. Caregivers assessed the way in which the doctors communicated the diagnosis as inhuman and not very sensitive (“He was 5 years old, 5 years old, there are doctors who suddenly tell you in a very inhuman way such as ‘Look! Your son has this’”; 27-year-old caregiver) because they concentrated on providing information on the changes in physical functioning that take place, the stages of the disease, and the death prognosis. Upon learning about the diagnosis, caregivers reported experiencing a stage of sadness and denial, and subsequently they resigned themselves to the diagnosis when investigating other sources such as the Internet.

When talking about the disease, they mentioned the age of diagnosis, the enormous pain caused by talking about the subject, the short life of their children, the frustration and

helplessness generated by knowing about the genetic factor inherent to the disease, and the deterioration of the quality of life of their loved ones brought about by the natural course of DMD.

Yes, it was very hard for me. I thought I would never be able to overcome it. No, no idea. Well, I had an uncle, but we always thought it was polio. We never thought it was something genetic and that muscular dystrophy could be passed from one generation to another... Moreover, now we have already learned that muscular dystrophy developed not only in our family but also in relatives of my grandmother” (38-year-old caregiver).

Meaning of the disease

When exploring the meanings that caregivers have about this disease, we found that they referred to DMD directly as “dystrophy” when they narrate their experiences about the

diagnosis and when they know the causes and development of the disease. But, in most statements, when talking about the disease, they focus on describing what the experience has meant to them and their children. When analyzing the responses of caregivers using the free association technique, we found that caregivers had difficulties in uttering the words despite the guidance received by the interviewer, and they preferred to construct narratives or explanations about the disease and what it meant to them.

Based on their responses, we created a

word cloud (Figure 2), and the word “they” was identified as the most frequently used word along with some other words that allowed them to articulate a narrative such as “because,” “then,” “when,” and “but”. When conducting a category analysis of the responses, caregivers described the disease with words such as: stubborn, hard, disaster, difficulty, the worst thing that ever happened to them, and suffering. They mentioned the implications of the social situations they had experienced: violation of rights, social lack of understanding, and invisibility of children.



Figure 2. Word cloud.

Simultaneously, they included aspects, such as blessing, social support, strength, and patience, in their discourse that had made them stronger. Finally, as key aspects in their narratives, caregivers referred to the concerns about their children and their role as caregivers as follows: disease process, agency capacity, and teaching how to defend themselves.

Living according to the needs of the boys and young men with DMD

This section describes the caregivers' experiences, their meanings, and the way in which they respond to the needs and experiences of their children, which progressively end up revolving exclusively around them. Therefore, there is a description of the tran-

sitions experienced by caregivers when assuming the role of a caregiver, the features of the relational experience with the boys and young men and healthcare personnel, and finally the experiences related to the social context.

Transitions in the assumption of a new role

The caregiving experience is characterized by continuous economic hardships. Being the caregiver in their families implicitly eased the transition involving changes at a personal, family, and social level to respond to the caregiving needs of their loved ones. On a personal level, some caregivers had to quit their jobs and seek other income sources as independent workers because most of them were the income providers for their families.

On a social level, caregiving implied that caregivers had to stay away from their friends because they assumed that they did not have any friends after the diagnosis of the disease and that they felt lonely. Therefore, they preferred to seek refuge in themselves or find religious support. In some cases, they reported that female friends, sisters, and mothers provided them with the most emotional and instrumental support. On a personal and family level, some caregivers remarked that they had separated from their partners; in other cases, they experienced deterioration in their intimate relationship with their partners as well as their relationship with their families. Caregivers with a partner expressed being constantly worried about them and the way in which they had taken charge of the disease of their children. They explained that, in some cases, their partners became irritable or isolated themselves from the process, which are answers that they justified because the caregivers were also experiencing their grieving process for the disease.

Experiences: relationships with the boys and young men with DMD

Positive personality traits of the boys were highly frequent in caregivers' descriptions of their kids, as well as emotional status and behaviors. Regarding personality traits, caregivers highlighted the traits related to sociability and strength to live with the disease. Caregivers constantly contrast traits regarded as negative with those of a positive trait (e.g., "He is bad tempered but sociable." — 27-year-old caregiver). Undesirable traits are justified by the characteristics of the disease and are considered "understandable".

The relational experience with the child presents positive and negative aspects. On a positive level, caregivers have built a bond characterized by trust and comradeship, expressed in a relationship they valued as loving, close, and pleasant, which allowed them to realize the needs of the boys and young men. On a negative level, caregivers felt discomfort when they moved away from the boy and reported occasional conflicts between family members generated tension.

The caregiver's experiences were oriented toward a constant willingness to act rather than reflect on their experiences. Therefore, their daily life was loaded with multiple tasks (such as moving the boy and addressing surgery complications) and imminent obligation to perform these tasks to meet the children's needs. This experience was physically and emotionally strenuous. On a physical level, caregivers reported that they "do not sleep well" and "feel tired" all the time. On an emotional level, the caregiver's role represents an "emotional burden" in which they did not express what they felt and, in some cases, ideas of suicide also appeared "It was very hard for me; there came a time when I even wanted to kill myself. I wanted to commit suicide and end the children's lives as well" (38-year-old caregiver).

Caregivers expressed continued concerns related to the loss of the boys' autonomy and

well-being and reported “seeing the deterioration of their children because of the disease” as a major difficulty. Therefore, the caregivers were worried about leaving the boys alone or not being present, about the boys not being able to fend for themselves, or about the boys getting sick or being unhappy.

Sometimes, caregivers perceived that they felt frustrated while managing some of the boys’ behaviors, mainly when the boys or young men did not express their needs and caregivers failed to realize these needs and faced the feeling of being unaware of the boys’ thoughts. Additionally, the moments of change in a boy’s mood and conflicts with their siblings are situations where they experienced special tension and impotence. The relationship of caregivers with boys and young men changes on the basis of their progress. In this regard, some caregivers expressed changes in the siblings’ relationship because of adolescence and concern for love relationships in which their children were involved.

However, at the level of our relationship, I do not find many difficulties. Maybe, currently, because he is in the onset of pre-adolescence and no longer likes to be called a child but rather a pre-teen. If I am going to kiss and spoil him, he says ‘Oh mummy, do not do that!’ He says that he is a pre-teen. Thereafter, I am clueless about how to handle the girlfriends’ issue because then he tells me ‘Mummy, will you give me a chocolate bar?’, then I ask him ‘What for?’ and he tells me that it is for a girlfriend, but he never talks or he feels embarrassed to talk about it. Therefore, I do not know how to handle this situation (36-year-old caregiver).

There are times when caregivers do not feel comfortable interacting with their children; they feel helpless, tired, and sad and, therefore, show emotional inhibition, fearing that the boys and young men would feel bad.

Experiences: relationships with healthcare personnel

The relational experience with healthcare personnel is somewhat ambiguous. The caregivers mentioned in their stories mainly experiences with doctors and, in some cases, mentioned physiotherapists and psychologists. On the one hand, caregivers feel that some doctors inspire trust and want to help them by providing informational support and having receptive, respectful, and sincere communication. In some cases, caregivers reported that boys have a “sincere” relationship in which they freely express their thoughts about their health status to their doctors; however, in some cases, boys are shy, and caregivers speak for them. However, doctors try to control the interactions to ensure that the boys and young men express their health status themselves.

On the other hand, caregivers reported negative experiences for themselves as well as their children in their interaction with healthcare personnel; this generated distrust and a perception of “not being interested in boys” and “not being concerned about their quality of life”. Sometimes, vertical communication was established, which generated discomfort in caregivers and its main outcome was silence despite the doubts they may have:

With regard to the doctors, I am a very calm person and therefore everything went well. Yes, I mean, I had my issues but all the same I did not go... I have never been able to tell them, I know who the doctor is, the one who gave me the diagnosis, the one who answered me like that, but then I am never going to go to ... to tell him, ‘Hey! Look! I think...’ I am not good at fighting anyone; I think ... I am one of those persons who ... and when I need authorizations, guardianships, etc., I go with a good attitude, and I always behave well without quarreling with anyone (27-year-old caregiver).

Caregivers mentioned that sometimes there were conflicts between the doctor and boys regarding the doctor’s style of communi-

cation or when there were doubts concerning the boys' report. These conflicts have generated a gap in the boy-healthcare personnel relationship and causes boys and young men to refuse physical therapies in some cases and avoid hospital contexts. In addition, in some cases, caregivers distrust the doctors' indications and consider that they do not have sufficient knowledge about the disease. Additionally, caregivers have faced humiliation by doctors and are questioned about their roles, thus resulting in a distant relationship in some cases, wherein the caregivers do not express their concerns about the process.

Experiences within the social context

The social context emerges as a continuous scenario of barriers for children as well as caregivers, as observed in the narratives of caregivers. Urban design, school context, health context, and behavior of other individuals are continuous sources of situations of violation of rights. Figure 1 shows the interrelationship of these scenarios and the main issues that were mentioned by the caregivers. The school with experiences of discrimination, the health context with an ambiguous relationship and the social context with situations of rights violations.

Caregivers believe that the city is not sufficiently fitted for boys and young men with DMD to be autonomous and move around easily. Assistance becomes necessary and the technology available is not enough to support boys and young men care. Besides, they regard social indifference as one of the main barriers.

In the narratives, schools appear as scenarios of continuous discrimination where abusive events by teachers and classmates were identified. Because of such school events, families discontinue the schooling of their children with DMD. In some cases, they have managed to get alternative school options to ensure that their children continue to be educated, but in most cases, these children experience a violation of their right to

education.

Within the context of healthcare, caregivers reported having experienced a constant struggle to obtain medical care for their children; they reported experiences of denial of resources for their children's health and long procedures to obtain authorizations for medical care. In the words of a caregiver:

... I filed a claim against the Health System in March or May, and they have not replied about the treatment for P3; therefore, the treatment has been stopped. Well, I try my best to continue his therapies because he has lost a lot of strength (36-year-old caregiver).

Finally, the discriminating images and remarks of other individuals close to or far from the life of boys and young men with DMD is concerning for caregivers. They believe that there is continuous disrespect and that they cannot always protect themselves; moreover, there are no mechanisms in place to enforce their rights. Caregivers have identified discrimination attitudes by other children and a continuous social evaluation that makes their children not want to go out because they do not feel comfortable when they are asked about their disease.

Rebuilding the meanings of the caregivers' experience

Adopting the role of a caregiver generates various changes in the life of caregivers. They have experienced several challenging situations and have felt frustrated and satisfied for the duty accomplished and for what their role has generated in them and their loved ones. Although they consider that the process itself has been devastating, caregivers continually reshape their experiences and try to become increasingly aware of their role and its implications; they develop strategies to support their children during the disease course and redefine their meaning of life by strengthening

their spirituality, the latter being the caregiver's "engine of life".

Caregivers expressed changes in their experience related to their interpretation of motherhood. One caregiver remarked that, although she has always been a good mother, "after the diagnosis I became a better woman, mother, and person". Other remarks emphasize the fact that they have stopped living their lives and now they live for their children, indicating that boys and young men with DMD are the new engine of their lives that brings them happiness.

When you are there, we cooperate and work for several things, and I have always been there! There is always 'that before and that after' phase because I value my children more and love them more, and suddenly, I want to spend more time with them; they have taught me to be a better woman and mother; they have taught me to be better in every way (38-year-old caregiver).

When handling difficult situations in the care of their children, some caregivers expressed that they always try to understand the situation their children are going through. When difficulties occur in communication, they foster conversations about the importance of respect in relationships and direct questions about the situation that generated the discussion and the expression of emotions in their children, thus trying to motivate them by suggesting alternative activities such as studying and working from home. When their children feel uncomfortable because of situations of social evaluation or discrimination, caregivers offer other perspectives on the situation and support them emotionally to control these difficult situations.

On a spiritual level, we identified resources that caregivers used to perform their role and make them stronger. First, they have learned about the management of the disease and accepted the fact that their children have to face this situation. Second, caregivers expressed

that they found solace in God to confront the disease. "But I say that God has given me the strength and resilience to continue providing care and be at my children's side every day."—38-year-old caregiver. Third, they have adjusted their lives to meet their children's needs by endowing the experience with a new meaning, as a blessing and an experience in which they have shown compassion toward their children throughout the process. Finally, they have re-built the meaning of life from their experience as a human being and mother, in which they see themselves from a positive self-image as caregivers.

Discussion

The purpose of this study was to analyze the meanings of being caregivers of boys and young men with DMD. As stated in the theoretical and empirical background on DMD and other health conditions, women—specifically mother—are the main caregivers (Nozoe et al., 2016, 2017). Generally, and according to García-Calvente et al. (2004), the findings of our study and the women justified the little involvement of men in the family, especially the father of the child. The caregivers' narratives indicate that a cognitive process of creation of meanings begins at the time of diagnosis, which is eventually transformed to allow coexistence with the disease, in which their main activity is to be caregivers. The meanings built on the experience of being a female caregiver are transformed in the progression of the disease (Samson et al., 2009). The study identified that in the initial stage the experience is assumed in a more catastrophic way, later a reconstruction of the meaning of life is evident in which the positive self-image as a caregiver becomes relevant (Figures 1 and 2). The analysis of these results led to the identification of three major topics around which the caregivers have built the sense of be caregivers of boys and young men who live with DMD: finding out about the disease, living according to the needs of

boys and young men, and providing a new meaning to life.

In regard to finding out about the disease, the results indicated that caregivers were informed of the diagnosis by the doctor, specifically regarding the course and nature of DMD. According to Dawson and Kristjanson (2003), receiving information from the physician is useful at all stages of the disease and not just at the time of diagnosis, which is when they were informed. Nevertheless, their need for information is surpassed by the need for a compassionate and “humanitarian” treatment by the doctor who conveys the information; however, none of the boys and young men received this treatment. Also, none of the participants in this study had received psychological support during their family member’s illness, which contradicts the approach of Landfeldt et al. (2016) and Bushby et al. (2010) according to which DMD requires multidisciplinary management.

These outcomes are evidence of the need for a psychosocial approach to DMD. These findings coincide with the literature on the treatment of the disease in which psychological accompaniment for patients and families is important to achieve a holistic approach aimed at the mental healthcare of caregivers of children with chronic diseases, as in the case of DMD (Birnkranz et al., 2018, Peay, Hollin, & Bridges, 2015). In fact, collaborative work between families and health professionals is essential to empower families and promote child self-care (Yang et al., 2018).

Denial was found to be a common coping strategy at the time of diagnosis owing to the high emotional impact caused by the prognosis received for DMD and, consequently, a way to mitigate it. Similar results have been reported by Thomas et al. (2014). In accordance with the statements of Dawson and Kristjanson (2003), the grieving process is a part of their daily lives for these women; this happens from the very moment of diagnosis and not only at the end of the course of DMD.

Finding out about the diagnosis of DMD is

the sign required to begin providing meaning to the disease in a context perceived as discriminatory, accompanied by a significant violation of rights and poor social support. These caregivers are mothers whose lives revolve around their children and for whom the impact of the diagnosis, which predicts the near death of their children, acquires a new dimension through a strongly binding relationship in which the daily and ineluctable care of the child becomes their main, and virtually the only, focus of life. This is only how the caregiving experience acquires meaning.

Caretakers of children with DMD embrace care as an affective act and perform it as a natural and moral duty in a responsible manner. In the words of Kleinman (2012), caring is a moral experience for these women because they are expected to take care of their children not only because they are women but also because they are mothers and are “responsible” for the development of the disease in their children (assuming that they have transmitted the DMD gene). In fact, qualitative research such as that of Samson et al. (2009) has revealed that, over time, parents of children with DMD gain confidence and the disease, now known, allows them to make sense of this painful experience and legitimize it, finding a coping option in serenity.

In this way, the disease eventually becomes the bond or connection between caregivers and their children/adolescents. The bond in this case, among others, is characterized by physical dependence (Moura et al., 2015). Genetics has imposed the need to assume a new role in these women of being the informal caregiver of a chronic boy or young man beyond being a mother or grandmother. According to Kleinman (2012), “being present” is an essential trait of caregivers despite having exhausted any alternatives for healing or performing any practical actions that could bring hope.

Without exception, caregivers reported feeling physically exhausted, and, with great difficulty, they admitted being emotionally

burned out. This result has been widely studied and reported in the literature (Gérain & Zech, 2018; López-Gil et al., 2009). As opposed to Landfeldt et al. (2018), none of the participants reported feeling clinically depressed.

As described by Thomas et al. (2014), the participants in this study also shared several coping strategies throughout the course of the disease, including positive re-interpretation (considering that this experience made them better mothers), active coping (searching for information on the Internet), and religious beliefs (God as a solace to face the challenges imposed by the disease). Thomas et al. (2014) have reported that emotional expression is often used as a coping strategy by caregivers of children with DMD. But, the findings of our study evince that this strategy is rarely used by caregivers.

With regard to living according to the needs of boys and young men, all the experiences of the caregivers accumulate until they substantiate the need to always be present with their child and “live for them”. Indeed, the progression of the disease increasingly demands care from caregivers and affects their quality of life. This new role implies radical changes in the life of caregivers such as the total or partial abandonment of their jobs; eventually caregiving becomes their job. In line with García-Calvente et al. (2004), caregiving is an informal job, with extreme demands on women’s capabilities which requires them to assume a new role in addition to those already under their responsibility (mother, daughter, psychologist, counselor, nurse, domestic worker, etc.). This lasts for endless days and affects different dimensions of their lives.

Living with a child with DMD involves an adjustment of personal/family life and family dynamics, as stated by Thomas et al. (2014). In accordance with the literature available, the women in our study shared several risk factors that diminish their quality of life and compromise their mental and physical health: sleep disorders (Nozoe et al., 2017); few social support networks and, consequently, deteriora-

tion of their social interaction (García-Calvente, 2004); and high levels of distress and stress associated with low resilience, low social support, and low income levels (Kenneson & Bobo, 2010).

In fact, a relevant element in the analysis of the caregivers’ narratives was the appearance of stress because of the economic hardships they have to face. As stated by Magliano et al. (2015) and Thomas et al. (2014) losing their jobs increases the psychological burden and decreases the income levels of caregivers, thus leading to financial hardship. When these women perform other jobs besides caregiving, they need to take time off work (Landfeldt et al., 2016), and the possibilities and opportunities to develop professionally are limited (García-Calvente et al., 2004).

Although death was a concern that emerged at certain moments in the narrative and caused anguish in mothers, caregivers chose to increasingly strengthen their relationship with their children. As opposed to Dawson and Kristjanson (2003), the participants in our study did not wish to talk to their children about death or the process of dying. Conversely, the topic of death is avoided to ensure that the boys and young men live in the present “here and now” and make the most of it. In fact, they disapprove of any member of the healthcare team talking to their children about death. This contradicts the findings of Abbott, Prescott, Forbes, Fraser, and Majumdar (2017), which state that talking about this issue is a need of the boys and young men and must be promoted by their family members.

Caregiving and the mother-child relationship are motivational components that transform and give meaning to life. As described by Magliano et al. (2014), it is an experience which provides significant benefits. In addition to this, relief of symptoms (pain, sleep disorders, and dyspnea) is an imminent need and is always present in caregivers, who fear the suffering and deterioration of their children. Taken together, these findings highlight the need that these women

have for their children to receive palliative care. Unfortunately, and as stated by Arias et al. (2011), our caregivers make little use of these services because they are unaware of their existence, and it is surprising that none of the children had been referred to this type of specialist.

The analysis of results and discussion of this study should be conducted within the framework of the recognition of their nature and scope and always avoid generalization of the results. DMD is an orphan disease and the number of boys and young men in Colombia is not known precisely although we know it is small; therefore, the sample size is considered relevant. In addition, the study can be reference for future research to influence decision making at the health and public policy level in Colombia. For example, offering a more holistic and integrated care system for patients and their families.

Regarding the scope of the study, we would like to highlight that this is a pioneering study in Colombia, which has allowed us to analyze the meaning of DMD for caregivers and eventually let their voices be heard and shared with the academic community. We hope that specific actions will be derived from it for the benefit of caregivers of boys and young men with DMD, which will promote respect for differences, inclusion, and visibility of boys and young men and caregivers.

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