ABSTRACT

Objective: To identify the main challenges of the family of a child with meningomyelocele (MMC) and correlate the Zarit Burden Interview (ZBI) scale with the World Health Organization Quality of Life-BREF (WHOQOL-BREF). Methods: Questionnaires were sent through an online platform to groups of parents and caregivers of children with MMC. Four WHOQOL-BREF domains were evaluated: physical, psychological, social and environmental. Results: All correlations among the domains of the WHOQOL-BREF questionnaire with the Zarit Burden Interview (ZBI) scale were negative and significant with a higher magnitude value for ZBI × psychological domain with −57.4% correlation (p < 0.001). Conclusion: The results of this study suggest the quality of life of mothers and caregivers of children with meningomyelocele is affected regarding functional capacity, emotional aspect, and mental health. Despite this, the participants present a positive view of their lives. The most common difficulties faced by caregivers were: performing bladder catheterization, providing general care, financial burden, and accessibility. Level of Evidence V, Expert opinion.

Keywords: Meningomyelocele. Neural Tube. Caregivers. Quality of Life.

INTRODUCTION

Meningomyelocele (MMC) is a malformation of the nervous system and is considered a complex defect in the neural tube closure. This change occurs in the fourth week of embryogenesis associated with frequent and severe neurological sequelae, which can cause significant morbidity and mortality. The prevalence of MMC in the world is one child per thousand live births; this is also the average incidence in the United States. Moreover, the lower the socio-economic development of the country, the higher the meningomyelocele index, as shown by the birth defects map published by the World Health Organization.2,3 There are several reports on MMC prevalence, in Brazil and in Latin America in the literature. Some of the data related to it are: 1.139/1,000 live births,4 5/1,000 1.2/1,000, an average of 1.01/1,000 live births.5 A study on the effect of folic acid before and after the prenatal process using a historical series of data from the state of

RESUMO

Objetivo: identificar os principais desafios da família de uma criança com Mielomeningocele (MMC) e correlacionar a escala de Zarit Burden Interview (ZBI) com o WHOQOL-BREF. Métodos: Foram enviados questionários via plataforma on-line para grupos de pais e cuidadores de crianças com MMC. Quatro dimensões foram avaliadas: física; psicológica; relações sociais; e meio ambiente do WHOQOL-BREF. Resultados: Todas as correlações entre os domínios do questionário WHOQOL-BREF com a escala de Zarit Burden Interview (ZBI) foram negativas e significativas com maior magnitude de valor para ZBI × domínio psicológico com −57,4% de correlação (p < 0,001). Conclusão: Os resultados desse estudo sugerem que a qualidade de vida das mães e cuidadores de crianças com MMC é afetada em relação a capacidade funcional, aspecto emocional e saúde mental. Apesar disso, apresentam uma visão positiva de sua vida. As dificuldades mais encontradas pelos cuidadores foram: realizar o cateterismo vesical; prestar cuidados gerais; financeira; sobrecarga de trabalho; e acessibilidade. Nível de Evidência V, Opinião do especialista.

Descritores: Meningomielocele. Tubo Neural. Cuidadores. Qualidade de Vida.
São Paulo, Brazil, found that MMC prevalence decreased by 35% from 0.57 to 0.37 per thousand live births after folic acid fortification.\textsuperscript{5,7} When dealing with a child with MMC needs, the family has the responsibility of providing basic care for nutrition, hygiene, stimulation to growth and development, as well as specific care resulting from organic dysfunction, such as urination problems, depending on the child’s neuromotor impairment. In order to properly treat and maintain the health of a child with MMC, there is a need for continuous follow-up with a multidisciplinary team consisting of pediatrician, neurosurgeon, orthopedist, urologist, geneticist, nephrologist, physical therapist, nurse, psychologist, and social worker, who must work in an integrated way, as the child needs to be assisted in various aspects.\textsuperscript{8}

The daily life of caregivers of children and adolescents with meningomyelecele at home is full of care and undergoes profound transformations within various emotional, economic and social aspects of the family.\textsuperscript{9} The approach to quality of life and burden of these caregivers, as well as their main difficulties, has not yet been addressed with relevance in the literature. The aim of this study is to identify the main challenges of the family of a child with meningomyelecele and their impact on the quality of life of these individuals.

**MATERIALS AND METHODS**

This is a cross-sectional study and was approved by the ethics committee of the institution. Questionnaires were sent through an online platform. The invitations were sent via Facebook to groups of parents and caregivers of children with meningomyelecele, who participated in websites and/or social networks about the subject (Table 1).

The questionnaires were answered by the person directly responsible for the care of the individual with MMC, with an informed consent form. Family caregivers of children diagnosed with MMC were included, aged 0 to 15 years. Data collection was performed through an online platform, directed and structured in two parts: the first, consisting of socio-demographic data involving the caregivers, through questions related to their age, gender, marital status, education, family income, knowledge about pathology and care provided; the second, through two instruments: the Zarit Burden Interview (ZBI) scale\textsuperscript{10} and WHOQOL-BREF.\textsuperscript{11} focused on the quality of life of caregivers and how they feel about their own quality of life, health, and other related domains.

The ZBI scale evaluated the caregiver’s burden through 22 questions subdivided into items on the caregiver-patient relationship, health condition, psychological well-being, finance, and social life. Searching for an instrument to assess the population’s quality of life, the World Health Organization (WHO) created a Quality of Life Study Group (WHOQOL Group), with the purpose of studying the concept and developing measurement instruments, from a cross-cultural perspective.\textsuperscript{12}

**Statistical analysis**

For the four WHOQOL-BREF dimensions evaluated: physical, psychological, social and environmental, descriptive statistics were generated such as average, standard deviation, coefficient of variation and minimum and maximum. In order to verify the reliability of the questionnaire among the variables each response level was converted into numbers. To assess the quality of the questionnaire applied, the conventional Cronbach alpha coefficient and the standard alpha coefficient were used. To correlate WHOQOL-BREF with the ZBI scale, Pearson’s correlation was used.

**RESULTS**

A total of 124 questionnaires were obtained through an online platform. The conventional Cronbach alpha for WHOQOL-BREF was 0.932 (n = 26 questions) and the standard alpha was 0.935 (n = 26 questions). For the ZBI scale, the conventional Cronbach alpha was 0.867 (n = 22 questions) and the standard alpha was 0.895 (n = 22 questions) (Table 2).

Regarding the participants’ profile, the average age was 34.96 ± 7.87, and 98.39% (n = 112) were female; 37.90% (n = 47) had high school + some high school; 47.68 (n = 59) had college + some college; 74.19% (n = 92) had their own housing; 68.55% (n = 85) were married; 31.45% (n = 39) had average income of 1.5 minimum wage, and 33.90% had income over three minimum wages. Practically two-thirds of the participants (n = 81) stated they had medium knowledge about meningomyelecele and 29% affirmed they had a lot of knowledge about the illness. 44.35% (n = 55) reported no difficulties with care.

In addition, the most common difficulties encountered by family caregivers were: bladder catheterization (16.94%, n = 21), general care (16.94%, n = 21), burden (13.71%, n = 17) and accessibility (13.71%, n = 17). A total of 39% of the participants had help from a specialist or health institution; 28.23% reported help from relatives; 4.84% from social networks; 3.23% had spiritual help. A quarter of the patients reported they had no help at all. A total of 72.58% of the homes (n = 90) were not visited by nurses. In the visits that did occur, topics discussed were how to perform catheterization (26.47%, n = 9), general children care (8.82%, n = 3) and vaccination (5.88% n = 2). However, 51.53% (n = 17) of caregivers stated that information provided by health professionals was not clear or did not correspond to reality. Regarding the self-referred participants’ quality of life mentioned in the WHOQOL-BREF questionnaire, 66% considered it to be good or very good (Figure 1).

| Table 1. Reliability statistics of World Health Organization Quality of Life-BREF questionnaire and Zarit Burden Interview scale. |
| Methodology | Cronbach alpha | Cronbach alpha based on standard items | N of items |
| WHOQOL-BREF | 0.932 | 0.935 | 26 |
| Zarit Burden Interview (ZBI) scale | 0.887 | 0.895 | 22 |

| Table 2. Descriptive statistics of the four domains of World Health Organization Quality of Life-BREF. |
| Domain | Average | Standard deviation | Coefficient of variation | Minimum | Maximum | Amplitude |
| Physical | 3.32 | 0.68 | 20% | 1.43 | 4.57 | 3.14 |
| Psychological | 3.39 | 0.71 | 21% | 1.50 | 4.67 | 3.17 |
| Social | 3.09 | 0.83 | 27% | 1.00 | 4.00 | 3.00 |
| Environmental | 3.01 | 0.63 | 21% | 1.38 | 4.13 | 2.75 |
| Overall average | 3.22 | 0.90 | 19% | 1.52 | 4.26 | 2.74 |
In terms of caregivers' health satisfaction, 45.16% (n = 56) of them reported being satisfied with their health and 4.84% (n = 6) were very satisfied. The domains of the WHOQOL-BREF questionnaire were divided into subdomains, and the domains with the lowest averages were social relationships (3) and environment (3.1). These domains presented Likert scale values below the overall average of 3.22. The subdomains with the lowest score were: participation and opportunities for recreational/leisure – environmental domain (2.5), financial resources – environmental domain (2.5) and sleep and rest – physical domain (2.9) (Figure 2). The domains with scale above the general average were the physical and psychological. The domain with the highest variability was "social relationships" (Table 3).

All correlation among the domains of the WHOQOL-BREF questionnaire with the ZBI scale were negative and significant with similar results to those found by Caro et al. in 2018 (Table 4).

Table 3. Correlation between World Health Organization Quality of Life-BREF domains with Zarit Burden Interview scale.

| Domains             | Physical | Psychological | Social  | Environmental | Total – Zarit Burden Interview (ZBI) scale |
|---------------------|----------|---------------|---------|---------------|--------------------------------------------|
| Correlation         | 1        | 0.727*        | 0.613*  | 0.681*        | – 0.567*                                   |
| P- value            | < 0.0001 | < 0.0001      | < 0.0001| < 0.0001      | < 0.0001                                   |
| N                   | 124      | 124           | 124     | 124           | 124                                        |
| Correlation         | 1        | 0.667*        | 0.676*  | – 0.574*      | – 0.574*                                   |
| P- value            | < 0.0001 | < 0.0001      | < 0.0001| < 0.0001      |                                             |
| N                   | 124      | 124           | 124     |               |                                             |
| Correlation         | 1        | 1             | 0.512*  | – 0.452*      | – 0.452*                                   |
| P- value            | < 0.0001 |               | < 0.0001|               |                                             |
| N                   | 124      | 124           |         |               |                                             |
| Correlation         | 1        | 1             | 1       | – 0.472*      | – 0.472*                                   |
| P- value            | < 0.0001 |               |         |               |                                             |
| N                   | 124      |               |         |               |                                             |
| Correlation         | 1        |               |         |               | – 0.567*                                   |
| P- value            | < 0.0001 |               |         |               |                                             |
| N                   | 124      |               |         |               |                                             |

*: Correlation is significant at level 0.01 (2 ends).

Table 4. Comparison between correlations found in Zarit Burden Interview scale and World Health Organization Quality of Life-BREF.

| Correlation               | Current study | Caro et al. (2008) |
|---------------------------|---------------|--------------------|
| ZBI x Physical domain     | – 0.567*      | – 0.394*           |
| ZBI x Psychological domain| – 0.574*      | – 0.282**          |
DISCUSSION

The study presented high internal consistency between responses since the alpha coefficients were 0.932 (WHOQOL-BREF) and 0.887 on the ZBI scale. In a study aimed to determine the strength of association between coping strategies and quality of life in adolescents with spina bifida using a KIDSCREEN – 52 questionnaire, an alpha of 0.93 was determined. In another study that evaluated the correlation between burden and quality of life of caregivers of children with meningomyelocele, the Informal Caregiver Burden Assessment Questionnaire (QASCI) was applied. The questionnaire evaluates the perception of burden in informal caregivers. In the WHOQOL-BREF questionnaire that also evaluates the perception of individuals’ quality of life, the Cronbach alpha was 0.88, showing good reliability. Quality of life implies different forms of understanding. According to Cipriano, quality of life is defined by the individuals’ feeling of well-being. These feelings can be measured objectively (income, employment, housing, material assets) and subjectively (security, privacy, recognition, affection, love, peace).

When considering the ZBI burden classes, we noticed that 51.61% (n = 64) of caregivers fall into the moderate burden category and 13.71% (n = 17) between the moderate to severe category. One-third on average had no burden 33.87% (n = 42), and less than 1% (n = 1) had severe burden (Figure 3).

| Correlation          | Current study | Caro et al. (2008) |
|----------------------|---------------|--------------------|
| ZBI × Social domain | −0.452*       | −0.040ns           |
| ZBI × Environmental domain | −0.472*   | −0.470**           |

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The literature reveals that mothers of children with neurogenic bladder report attrition due to the burden or supervision required, leading to physical, psychic and social problems in their own lives. Some factors may interfere with the quality of life of caregivers of patients with MMC, such as socioeconomic status, injury site, locomotive difficulties and chronic illness associated with the caregiver. The long period spent and high cost involving care lead to financial difficulties. The visits of nursing professionals at the participants’ homes are insufficient since 72.58% do not have frequent visits by professionals and when they do, it is generally to provide information on bladder catheterization. The absence of health professionals in the homes of children with MMC is a known issue. Unlike many health professionals, families often do not find catheterization simple. Many caregivers describe this procedure as something that binds them with the child, due to the need of the procedure to be performed several times a day. The quality of life profile for family caregivers of children with MMC was considered good and very good, totaling 65.31% considering the sum of both cases (Figure 1). The search for guidance and support in the process of caring for their children is a valuable resource in coping with their child’s condition, as well as with other people in society. The initial denial of the illness and revolt in front of the situation changes slowly with the acquisition of information. This information can be acquired through contact with other mothers, whose children live in the same situation. The main difficulty in caring for children with meningomyelocele is bladder catheterization. Thus, the families of these children also learn to live with prejudices and stigmas related to motor difficulties and urinary and fecal incontinence. Both interfere with social activities. In a study investigating the thoughts and feelings of family caregivers about performing daily care in children with MMC (such as catheterization), it was suggested there should be physician participation and even the need for a psychologist to work with the interdisciplinary team, so these professionals can manage emotional difficulties with everyone involved in this exhausting procedure.

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Figure 3. ZBI burden classes.

The domains and subdomains measured on the WHOQOL-BREF scale presented worse values for the environmental domain and the subdomains participation and opportunities in recreation and financial resources (Figure 2). This is probably because the caregiver is deprived of the means for leisure. The WHOQOL-BREF method was developed collaboratively and simultaneously in several centers, including countries with different levels of industrialization, availability of health services, importance
of the family and dominant religions, among others, in addition to using an interactive data input among the researchers, at each stage of their development, allowing constant influence in the process. The results found by Caro et al. (2018) aimed to identify the level of burden and quality of life of family caregivers of stroke patients and to investigate the correlation between burden and quality of life (including physical, social, psychological and environmental domains). Therefore, different correlations can be compared; the physical and environmental domains had negative and significant correlations in both studies.

On the burden index using the ZBI scale methodology the average score ± standard deviation was 26.93 ± 13.09 (Figure 3). In a study conducted in two institutions where two different groups were evaluated (Group 1 with clean intermittent catheterization and Group 2 with use of diapers), the averages on burden were respectively 44.71 ± 10.92 and 22.94 ± 12.22, showing a significant difference between groups (p = 0.01). The higher score of Group 1 can be explained by the fact that bladder catheterization takes longer to be executed, since the procedure needs to be standard at specific time intervals, and dealt with caution, as it is a clean and non-sterile technique designed to avoid any sort of infection or complications arising from the introduction of a catheter through the urethra. Subjectivity deals with human feelings, i.e., the qualitative perceptions of the experiences lived. Despite the difficulties presented, caregivers remain content with their quality of life and continue to break paradigms, facing often insurmountable barriers and challenges (Figure 4). Many different problems may bring suffering and despair as caregivers know that treatment is a continuous process to improve the development and life of the child.

CONCLUSION

The results of this study suggest that the quality of life of caregivers of children with meningomyelocele is affected regarding functional capacity, emotional aspect, and mental health. Despite this, the participants present a positive view of their lives. This ability to overlook problems and face difficulties is a striking feature of these families and should encourage programs to improve care and information for caregivers of children with meningomyelocele.

The most common difficulties encountered by caregivers were performing bladder catheterization, providing general care, financial burden, and accessibility; however, even with these difficulties, they constantly find strength and correlate the welfare of the child to their own well-being.

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