

QUALITY OF LIFE AND CHALLENGES OF FAMILY MEMBERS OF CHILDREN WITH MENINGOMYELOCELE

QUALIDADE DE VIDA E DESAFIOS DE FAMILIARES DE CRIANÇAS COM MIELOMENINGOCELE

RODRIGO SANTOS BUORO¹ , MONICA PASCHOAL NOGUEIRA¹ 

1. Instituto de Assistência Médica ao Servidor Público Estadual (IAMSPE), São Paulo, SP, Brazil.

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.

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: Meningomielocelo. Tubo Neural. Cuidadores. Qualidade de Vida.

Citation: Buoro RS, Nogueira MP. Quality of life and challenges of family members of children with meningomyelocele. Acta Ortop Bras. [online]. 2020;28(6):291-295. Available from URL: <http://www.scielo.br/aob>.

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.¹

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 pre-natal process using a historical series of data from the state of

All authors declare no potential conflict of interest related to this article.

The study was conducted at Instituto de Assistência Médica ao Servidor Público Estadual.

Correspondence: Monica Paschoal Nogueira. Avenida dos Maracatins, 426, Moema, São Paulo, SP, Brazil, 04089000. monipn@uol.com.br

Article received on 06/05/2020, approved on 01/07/2020.



São Paulo, Brazil, found that MMC prevalence decreased by 35% from 0.57 to 0.37 per thousand live births after folic acid fortification.^{6,7} 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.⁸

The daily life of caregivers of children and adolescents with meningomyelocele at home is full of care and undergoes profound transformations within various emotional, economic and social aspects of the family.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 meningomyelocele 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 meningomyelocele, who participated in websites and/or social networks about the subject (Table 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

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) scale¹⁰ and WHOQOL-BREF,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.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 0.887 (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 and 47.58 (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 meningomyelocele 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 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

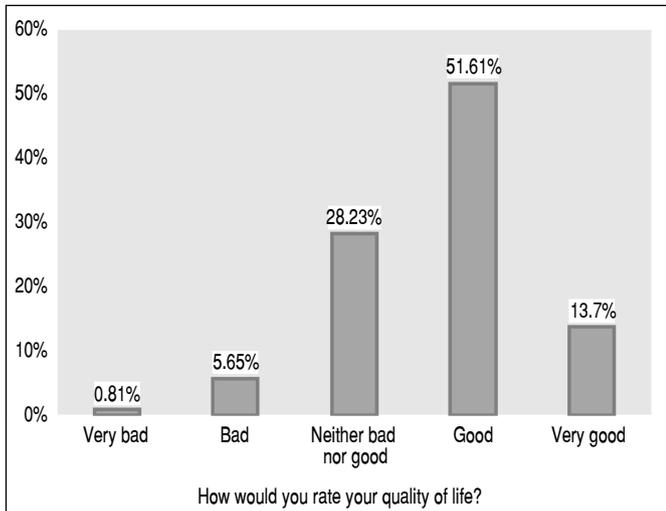


Figure 1. Quality of life profile of family members and caregivers of children with meningomyelocele.

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)

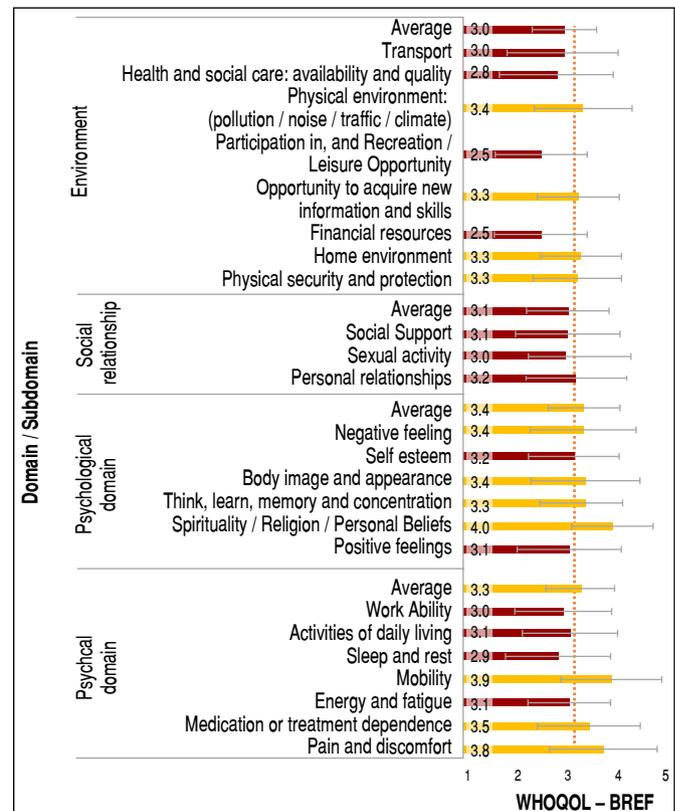


Figure 2. Domains and subdomains measured on WHOQOL-BREF scale.

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

*: 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 × Physical domain	- 0.567**	- 0.394*
ZBI × Psychological domain	- 0.574**	- 0.282 ^{ns}

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 × Social domain	-0.452 ^{**}	-0.040 ^{ns}
ZBI × Environmental domain	-0.472 ^{**}	-0.470 ^{**}

ns: not significant; **: significant at 1% by t-test; *: significant at 5% by t-test.

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).

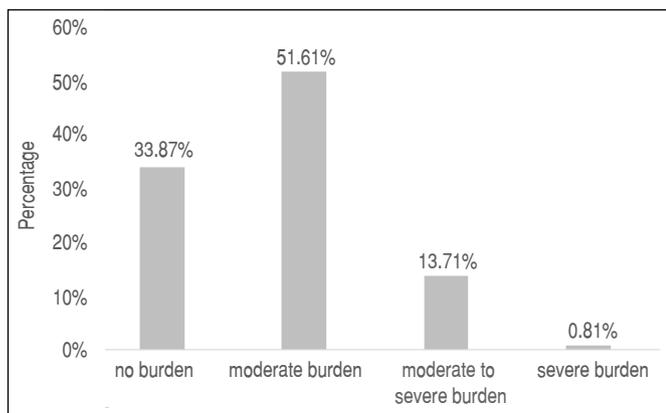


Figure 3. ZBI burden classes.

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 obtained an alpha of 0.93.¹³ 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.¹⁴

Quality of life implies different forms of understanding. According to Cipriano,¹⁵ 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).

On average, 98.39% of responding caregivers were women (Figure 1), as well as 87.5% of volunteers from forty caregivers of patients with MMC recruited in two institutions. The predominance of women is in line with what was found in the literature, where the tradition of women is emphasized in assuming the primary responsibility of caring for children, the older population and the bedridden.¹⁴ It is emphasized that the mother was the main representative in the monitoring and care of these children as they were present in all consultations and other forms of care.

Figure 3 shows that only 5.65% had little knowledge of MMC. Although the mothers presented little or no knowledge about the child's condition until the time of diagnosis, we found that after the initial shock of the news about their child having the condition some of the mothers were in search for information about the

illness. 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.¹⁶

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.¹⁷

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.¹⁸ The long period spent and high cost involving care lead to financial difficulties.¹⁹ 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 (Figure 2). 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.¹⁶

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. Attempting to preserve and care for the quality of life of their children, mothers put themselves behind by neglecting aspects such as vitality, mental and emotional health; as such, these results show us that the multidisciplinary team must also extend care to the families so they express their fears, anguish, and yearnings arising from physical and emotional weariness.¹⁸ In this study, the results found were different from those reported by the authors cited.

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.¹² 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.²⁰

On the burden index using the ZBI scale methodology the average score \pm 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.¹⁵

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).¹ 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.

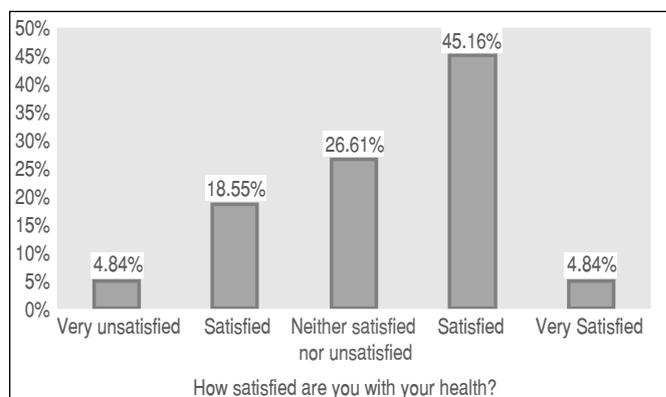


Figure 4. Profile related to health satisfaction of family members and caregivers of children with meningomyelocele.

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.

AUTHORS' CONTRIBUTIONS: Each author contributed individually and significantly to the development of this article. RSB: writing of the article, data analysis, interpretation, participation in the review process and approval of the final version of the manuscript to be published; MPN: design and planning of the activities that led to the study, study orientation, critical review of the intellectual content, and approval of the final version of the manuscript to be published.

REFERENCES

- Nejat F, Kazmi SS, Habibi Z, Tajik P, Shahrivar Z. Intelligence quotient in children with meningomyeloceles: a case-control study. *J Neurosurg.* 2007;106(2):106-10.
- Grillo E, Silva RJM. Neural tube defects and congenital hydrocephalus. Why is prevalence important? *J Pediatr.* 2003;79(2):105-6.
- World Health Organization. World Atlas of Birth Defects [Internet]; Geneva; 2003 [accessed on 2020 Aug 13]. Available from: <https://bit.ly/30XNLuN>.
- Nazer JH, López-Camelo J, Castilla EE. Results of thirty years of epidemiological surveillance of neural tube defects. *Rev Med Chil.* 2001;129(5):531-9.
- Bevilacqua NS, Pedreira DA. Fetoscopy for meningomyelocele repair: past, present and future. *Einstein.* 2015;13(2):283-9.
- Teixeira E, Oliveira MC, Sauron FN, Santos LSB. Terapia ocupacional na reabilitação física. São Paulo: Roca; 2003.
- Freitas GL, Sena RR, Faleiros e Silva JC, Castro FFS. Rehabilitation of children and adolescents with myelomeningocele: the daily life of mothers-caregivers. *Rev Gaucha Enferm.* 2016;37(4):145-57.
- Mitchell LE, Adzick NS, Melchionne J, Pasquariello PS, Sutton LN, Whitehead AS. Spina bifida. *Lancet.* 2004;364(9448):1885-95.
- Ministério da Saúde. Agência Nacional de Vigilância Sanitária (BR). Resolução de Diretoria Colegiada – RDC nº 344, de dezembro de 2002 [Internet]. Brasília (DF); 2010 [accessed on 2020 Aug 13]. Available from: <https://bit.ly/30Tsr9F>.
- Zarit SH, Reeve KE, Bach-Peterson J. Relatives of the impaired elderly: correlates of feelings of burden. *Gerontologist.* 1980;20(6):649-55.
- Fleck MP. The World Health Organization instrument to evaluate quality of life (WHOQOL-100): characteristics and perspectives. *Cienc Saude Colet.* 2000;5(1):33-8.
- Oliveira MF, Silva LM, Oliveira F, Gomes TT. Quality of life of caregivers of patients with cerebral palsy. *J Health Sci Inst.* 2013;31(4):421-4.
- Kós RS, Ricetto CLZ, D'Ancona CAL. Impact of urological treatment on quality of life of children with myelomeningocele and burden on their caregivers. *Rev Estima.* 2016;14(2): 84-90.
- Rosas AS, Yangua RP. Compreendendo el afrontamiento y la calidad de vida en adolescentes con espina bífida. *Casus.* 2017;2(3):139-46.
- Cipriano MAB, Queiroz MVO. Care with a child porter of meningomyelocele: family living. *Rev Rene.* 2008;9(4):72-81.
- Morais DM. Qualidade de vida relacionada à saúde em mães de crianças e adolescentes com mielomeningocele [dissertation]. Uberlândia: Universidade Federal de Uberlândia; 2010.
- Zarit S, Zarit J. Instructions for the burden interview. University Park: Pennsylvania State University; 1987.
- Marques MG, Verza A, Rossatti KZ, Carbonero FC, Campos D. Quality of life of the children caregivers with neurological dysfunction. *Cad Pos-Grad Disturb Desenvol.* 2016;16(2):16-23.
- Costa EC, Veloso RD, Feitosa JJ. Children with hydrocephalus: difficulties and mother's experience. *Rev Interd.* 2013;6:71-9.
- Caro CC, Costa JD, Cruz DMC. Burden and quality of life of family caregivers of stroke patients. *Occup Ther Health Care.* 2018;32(2):154-71.