

Commentary: "Quality of Life in Caregivers of Children and Adolescents with Osteogenesis Imperfecta"

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Background

Any treatment approach involving children must take into account their families. In cases of chronic childhood diseases, the family plays an important role in the process of caring for the child¹.

Families with genetic diseases affecting one of their members frequently report a distressing delay in receiving the diagnosis^{2,3}. After the diagnosis, they often have to face the lack of training and knowledge of health professionals regarding the disease as well as the weaknesses of health services³⁻⁵.

Chronic childhood diseases have physical, social, and psychological impacts on family members. Not only must they perform their daily duties, roles, and obligations but also meet the specific needs resulting from the child's disease. This causes severe disruption in the family dynamics⁶. Oftentimes, nevertheless, only one member of the family assumes the role of "primary caregiver," usually the mother. When this occurs, a common effect is the mother's physical and psychological exhaustion due to the accumulation of functions within the home¹.

As a result of this routine, the caregiver often develops a health problem and can be considered a "hidden patient"⁷. One strategy to evaluate the impact of the child's disease on the caregiver is performing an assessment of the quality of life (QOL). QOL is based on the following aspects: subjectivity, multidimensionality, and presence of positive (i.e., mobility) and negative (i.e., pain) dimensions⁸.

Our results

In the study entitled "Quality of life in caregivers of children and adolescents with Osteogenesis Imperfecta,"⁹ we conducted a cross-sectional assessment of QOL in caregivers of patients with osteogenesis imperfecta (OI) using the abbreviated version of the World Health Organization Quality of Life questionnaire (WHOQOL-BREF)¹⁰, a generic instrument that has been validated to Portuguese language¹¹. The WHOQOL-BREF containing 26 questions distributed in four QOL domains: Physical, Psychological, Social, and Environmental. In this study, caregivers were defined as individuals who had direct contact with children or adolescents with OI (care recipients) and were in charge of coordinating and providing for the recipients' basic needs. Twenty-four caregivers

from 24 unrelated families were included in the study. Their mean age was 39 ± 9.1 years, and two caregivers had OI (types I and IV, respectively) but no major physical limitation. Eighteen (75%) caregivers were female and 16 of them were mothers of children with OI. These 24 caregivers were responsible for the care of 27 patients with OI. Our data suggest that the majority of caregivers of patients with OI are their mothers, in agreement with a previous study showing that mothers were caregivers of 80% of children living with chronic disease¹².

Mothers are generally more involved than fathers in the disease management process; they usually act as chaperones during their children's hospital visits and are more likely to interact with care management teams¹. In our study, 13 care recipients had type IV, 10 had type I, and four had type III OI. WHOQOL-BREF scores were highest for the social relationships domain (15.2 ± 3.7) and lowest for the environment domain (12.9 ± 2.9). The distribution of WHOQOL-BREF scores was not associated with the number of fractures of care recipients (t-test: physical health, $p = 0.67$; psychological health, $p = 0.25$; social relationships, $p = 0.94$; environment, $p = 0.66$; overall, $p = 0.86$), OI type, or caregivers' socioeconomic status. The domains Physical ($p=0.002$), Psychological ($p=0.003$) and Environmental ($p=0.004$) were significantly lower than those of healthy individuals from the original instrument validation sample¹⁰. These differences persisted after exclusion of the two caregivers with OI from analyses.

Our participants showed lowest WHOQOL-BREF scores for the environment domain, which assesses respondents' financial resources, physical safety and security, home environment, opportunities to acquire new information and skills, participation in and opportunities for recreation/leisure, and physical environment (pollution, noise, traffic, climate), as well as the accessibility and quality of health and social care and transportation⁹. WHOQOL-BREF environment domain scores were also lowest in other studies, which may be related to socioeconomic issues in Brazil^{11,13-15} or may be attributed to having children with OI.

Future perspectives

In our study assessing QOL through the WHOQOL-BREF questionnaire, the results of caregivers of patients with OI were significantly different from those of the general Brazilian population, with lower scores for physical health, psychological health, and environment domains. This indicates that, in addition to treating and caring for children and adolescents, health professionals

should observe, listen to, and care for caregivers within their specific contexts as they may also become patients. A caregiver with poor health may not be able to provide adequate care for the child.

We emphasize the importance of carrying out new studies with larger sample size with comparison of OI caregivers versus other caregivers. Also, the conduct of qualitative studies with this population is important to identify the possible factors that have contributed to these results.

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