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## Quality of life of caregivers of children with visual impairment: A qualitative approach

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### ABSTRACT

**Background:** Receiving a diagnosis of a child with untreatable visual impairment (VI) may have a negative impact on parents and caregivers, and affect their quality of life (QoL).

**Aims:** To use a qualitative research approach to determine the impact that caregiving a child with a VI has on the QoL of caregivers in Catalonia (Spain).

**Methods:** An observational study was designed in which nine parents of children with VI (6 mothers) were recruited following an intentional sampling scheme. In-depth interviews were conducted, and a thematic analysis was performed to identify main themes and subthemes. The QoL domains defined in the questionnaire WHOQoL-BREF guided data interpretation.

**Results:** An overarching theme was defined (*the weight on one's shoulders*), as well as two main themes (*obstacles race* and *emotional impact*) and seven subthemes. QoL was negatively affected by a general lack of knowledge and understanding regarding VI in children and its implications for children and caregivers, whereas social support, gaining knowledge, or cognitive reappraisal had a positive effect.

**Conclusions:** Caregiving for children with VI affects all QoL domains, resulting in persistent psychological distress. Both administrations and health care providers are encouraged to develop strategies to assist caregivers in their demanding roles.

### What this paper adds?

A qualitative research was conducted to collect first-person testimonies about the experience of caregiving children with visual impairment. This approach led to an analysis of the confluence of various themes, which could be transferred into quality of life (QoL) domains, and to examine their changes over time. Thematic analysis identified several factors that heighten the disability of children beyond the actual sensory limitation, thus hindering their potential and having a negative impact on their QoL and on that of their caregivers. Elements that either hinder or assist care were explored and their role as barriers or benefits on the QoL of caregivers was determined.

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## 1. Introduction

The International Classification of Diseases 11th Revision (ICD-11, WHO, 2020) describes three categories of visual impairment (mild, moderate, severe) and three categories of blindness (from light perception or counting fingers to no light perception); it also encompasses an additional category for undetermined or unspecified visual impairment (VI). Overall, the prevalence of moderate or severe VI and blindness in children (0–18 years old) has been estimated at 6.5 cases per 10000, although studies report a wide range of values and aetiologies (Bezabih et al., 2017; Gyawali et al., 2017; Hatton et al., 2013; Haugen et al., 2016; Rahi & Cable, 2003; Resnikoff et al., 2004; Solebo & Rahi, 2014; Solebo et al., 2017; WHO, 2021). VI in children is often associated with a rare disease, which, given its low prevalence, commonly entails a limited medical understanding of the condition and a reduced or almost absent public awareness of VI itself and its implications on children development. This may restrict their social inclusion by means of hindering both their activity and participation in society.

In Spain as a whole, and in Catalonia in particular, prevalence of VI in children has been documented at about 6 cases per 10,000 (IMSERSO, 2022), with about 800 cases in Catalonia (Catalunya & d'Educació, 2021). Of these, 95% of children are enrolled in ordinary schools, with only a minority joining special education institutions. The Department of Education of the Catalan Government ensures the inclusion of students needing specific educational support, coordinating the actions of psychopedagogical counseling and guidance teams with the ONCE (Spanish Organization for the Blind) Educational Resource Center for the Visually Impaired.

Literature on caregiving children with chronic conditions including VI (Lupón et al., 2018), describes the associated impact on parents and other caregivers in terms of stress, worries, burden and work and economic related implications, amongst others (Al Qadire et al., 2020; Commissariat et al., 2020; Picardi et al., 2018; Zaidman-Zait et al., 2016). Parents of children with rare diseases also often report feeling alone and lost facing their situation (Baumbusch et al., 2019; Cardinali et al., 2019; Currie & Szabo, 2018; Currie & Szabo, 2019), with a negative impact on their quality of life (QoL).

The World Health Organization (WHO) describes QoL as “an individual’s perception of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns” (WHO, 1998). Four domains of QoL were considered in the questionnaire WHOQOL-BREF, which is the short version of WHOQOL-100 (WHO, 2012): *physical health* (energy and fatigue, sleep and rest, etc.), *psychological* (negative or positive feelings, memory, concentration, etc.), *social relationships* (personal relationships, social support, etc.), and *environment* (financial resources, health and social care accessibility and quality, etc.). The goals, expectations, standards and concerns that parents had before their child is diagnosed with incurable VI may be transformed, and in the corresponding adjustment process, one if not all domains of QoL can be affected.

In Catalonia, qualitative research has never been conducted on the implications that informal care of children with VI have on the QoL of their parents and caregivers. The analysis of in-depth interviews provides information on the experiences of caregivers from an ecological viewpoint, progressing from an individual and intimate sphere to a wider perspective, and it reflects the temporal evolution of these experiences. This approach may determine how caregivers interact with other people and contexts involved in childcare (family, friends, teachers, health system, school, administration, etc.) and their mutual influence, and how this interaction influences their caregiving experience and their QoL.

The aim of the present research was to provide a qualitative approach of the experience of caregiving a child with VI in Catalonia, and to determine the possible repercussions on the QoL of caregivers by considering the actual context and the government policies and regulations regarding disability, schooling, social services and health.

## 2. Material and methods

### 2.1. Study design and participants

A qualitative observational study was conducted through in-depth interviews with caregivers of children with VI. The target population was that of caregivers of children with VI residing in Catalonia, including mothers, fathers and legal guardians who understood and expressed themselves fluently in either Catalan or Spanish.<sup>1</sup> Occasional caregivers (e.g., grandparents, babysitters, etc.), parents or legal guardians with comprehension or communication difficulties in Catalan or Spanish, those of adults with VI and those of children with a level of VI not contemplated as such in the ICD-11, were excluded from the study. Sampling was intentional and several associations and health professionals in the field of VI acted as intermediaries. This research received the approval of the Ethical Review Board of the *Hospital Universitari Mútua de Terrassa* (02/2018) and all participants signed an informed consent after the aims of the study were explained to them. Data collection was conducted ensuring the anonymity of the participants in the study.

### 2.2. Procedure

Candidates were contacted via email or telephone to schedule the date, time and location to conduct the interview. All interviews took place between March 2018 and March 2019. Interviews were recorded with a mobile phone (Samsung Galaxy J3 2016) and the AUDIPO: Audio Speed Changer app (Google Play Store) was used while performing textual transcriptions.

Meetings with participants were structured in two parts. Firstly, participants were informed of the objectives of the study and were

<sup>1</sup> Spanish is the official language of the entire Spanish territory; In Catalonia, Catalan is also the official language. The authors understand and speak both languages fluently.

asked to sign an informed consent. Sociodemographic data of the family nucleus was collected, as well as data on VI and on the use of information and communication technologies (ICT) or Internet in relation to the visual condition of the child. Secondly, the actual interview took place. The following starting remarks were shared: “Could you tell me how you live (and have lived) the experience of being a mother/father of a child with VI? If you think this has had an impact on various areas of your life, could you explain how? It is not necessary to follow any order; this is a totally open interview format”. This approach aimed at not only focussing on an aspect or moment in time, but to allow stories to flow and develop. A posterior inductive analysis was used to determine main themes, sub-themes, the relationship between them, and the perception of temporal changes.

A single meeting per participant was conducted, with a duration ranging from 1 h 45 min to 2 h 15 min.

### 2.3. Data analysis

The transcribed texts of the recorded interviews were submitted to a thematic analysis following the guidelines of [Braun and Clarke \(2006\)](#). In brief, thematic analysis is a six-phase iterative process aiming at the identification and analysis of patterns (themes) in a data set. This process was conducted through in-depth discussion amongst the authors until consensus was reached. The phases of the analysis were:

- *Phase 1. Familiarizing yourself with your data:* transcriptions of all interviews were revised for correction ( $8471 \pm 1597$  words per interview, mean  $\pm$  standard deviation).
- *Phase 2. Generating initial codes:* a list of more than 60 codes and subcodes was generated (e.g., uncertainty, making decisions, guilt, gender role, etc.).
- *Phase 3. Searching for themes:* a revision and analysis of segments of texts and corresponding interrelated codes resulted in a first draft of 9 themes (e.g., control, discomfort expression, diagnosis, etc.).
- *Phase 4. Reviewing themes:* posterior review iterations of themes, subthemes and segments of text led to a reduction in the number of themes from 9 to 4.
- *Phase 5. Defining and naming themes:* the approach “define and refine” described by [Braun and Clarke \(2006\)](#) led to the suggestion and definition of two main themes and one core theme for analysis, as described in the Results section below.
- *Phase 6. Producing the report:* the outcome as described in the Results and Discussion sections below correspond to this phase.

The four QoL domains of the WHOQoL-BREF questionnaire (*physical health, psychological, social relationships, and environment*) were considered to facilitate data interpretation and provide a comprehensive understanding of the impact of caring for children with VI, on the quality of life of their caregivers.

## 3. Results

### 3.1. Study sample description

Nine participants were included in the study (6 mothers and 3 fathers). [Table 1](#) provides a summary of the characteristics of the sample (including demographic data and other relevant information).

At the time of the interview, none of the mothers working part-time jobs ( $n = 2$ ) considered increasing working hours. Of those working full-time jobs ( $n = 4$ ), two of them had flexible working hours and two of them had been working part-time until recently. One mother working full-time had recently lost three jobs due to the impossibility of maintaining flexible working hours. All job-related changes were associated with the caregiving burden and its implications (family management, administrative procedures, medical visits, etc.).

Regarding online search for information, all parents would have appreciated guidance from professionals, as this search required too much of their time and they were not able to assess the quality of the information they found, which could be very simple and banal or use a language too scientific and technical for them to understand. Most of them had discussed the information found online with

**Table 1**  
Characteristics of the study sample.

|  | Mothers (n = 6)  | Fathers (n = 3)                       |
|--|--|---------------------------------------|
| AGE (mean $\pm$ SD) [range], years             | 45.7 $\pm$ 7.3 [40–56]   | 47.7 $\pm$ 4.5 [43–52]                |
| EDUCATION LEVEL                                | Secondary (n = 2); University (n = 4)  | Secondary (n = 2); University (n = 1) |
| OCCUPATION                                     | Full-time (n = 4); Part-time (n = 2)   | Full-time (n = 3)                     |
| FAMILY STRUCTURE                               | Nuclear family: mother and father living with children (n = 9)   |                                       |
| USE OF ICT RELATED TO VI (tools and frequency) | Familiarity with ICT before the onset of VI (n = 9); Use of computers and smartphones to search information related to VI (n = 9); Frequency of use related to online search of information related to VI reported as ‘often’ or ‘very often’ (n = 8). |                                       |
| (reported usefulness)                          | ICTs are rated as useful in relation to the VI of the children (n = 9); Web search was conducted on their own initiative (n = 9); It would be positively valued if health professionals suggested useful links (n = 9).                                |                                       |

SD: Standard deviation; ICT: Information and communication technologies; VI: visual impairment

their health care professionals, but they reported poor feedback from them, as well as a skeptical and reluctant predisposition to address this issue.

Related children with VI were 4 girls and 5 boys, with ages between 6 and 18 years ( $12.6 \pm 4.5$  years). All of them were enrolled in mainstream schools and attended primary ( $n = 4$ ), secondary ( $n = 4$ ) and university ( $n = 1$ ) education. In two cases, there was no clear diagnosis of the condition and in the rest of the cases the diagnosis was known for at least five years, except in one case that was diagnosed the previous year. There were four children with comorbid conditions and two were siblings.

### 3.2. Thematic analysis report

The approach “define and refine” described by Braun and Clarke (2006) led to the proposal of two main themes for analysis: *obstacles race* (i.e., extrinsic factors that could act as barriers or impediments both in the full development of children and in the caregiving process), and *emotional impact* (i.e., parents’ intrinsic factors related with their skills to manage the situation). Both main themes influence each other and form a core or overarching theme, which was defined as *the weight on one’s shoulders* to reflect the permanent responsibility all participants manifested feeling, regardless of factors such as age (theirs and of their children), severity of VI, time since diagnosis, etc. (Fig. 1).

Each main theme was built upon several subthemes, described next and illustrated with specific remarks from parents (masculine forms are used in order to prevent any identification of participants and children).

#### 3.2.1. Obstacles race subthemes

**3.2.1.1. (Un)knowledge regarding Visual Impairment.** This subtheme refers to the general lack of knowledge about VI and its causes, its influence in child development opportunities, and its contribution to harden the *obstacle race* of caregivers.

Parents confessed their initial ignorance: they were completely unaware of the medical condition, they failed to notice signs and clues of visual problems, and they minimized the situation and the repercussions for the overall development of the child.

[Participant 1] “Since he was very young, he had the habit of watching television with his head turned sideways.... I knew there was a problem, but I did not realize its severity.”

In those cases in which health professionals did not possess enough knowledge of the condition, or were not sufficiently alert to the signs, diagnosis was delayed, increasing the uncertainty and concern of caregivers.

[Participant 9] “It had been a couple of years since visual acuity was worsening, but since a little brother was born at that time, he was thought to be a malingerer... and six years elapsed! Once you reach a diagnosis it is like a liberation, despite the upheaval.”

Some parents expressed a lack of understanding by teachers of the complexity of the VI situation, who acted with disbelief and/or passivity.

[Participant 5] “The teacher told me: sometimes I do not notice he has low vision, he sees such fine details... a dark spot on a white table. Until this year after school camp I did not realize he did not see a thing, he was falling over all of the time!”

All participants noted that VI was something very difficult to know and comprehend, and was considered a real barrier for parents.

**3.2.1.2. Enjoy proper support.** This subtheme describes the relationship of parents with other people or institutions acting as potential sources of support, which may be provided or negated, with a different impact on caregiving.

All parents noted a general coldness and lack of sensitivity of health personnel at the time of the diagnosis, and the fact that they often were discredited for their concerns towards the welfare of the child.

[Participant 8] “A year ago we had been through the same situation with a brother, and they told us simply: you know what this is about. We couldn’t articulate a word and just left.” [Participant 5] “I think it was the hardest moment, when you see something that nobody else sees... are

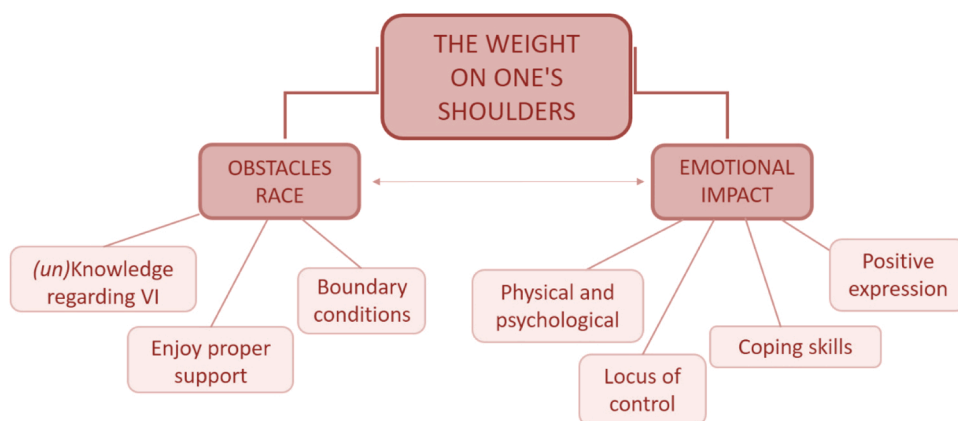


Fig. 1. Thematic map of themes and subthemes.

you crazy?. they told me I was imagining things!”.

After the diagnosis, professional politeness was valued, but support was still considered insufficient, and not adequate to respond to the needs and expectations of parents, and the signs of empathy were scarce. For those parents attending mental health specialists for support, their assistance was considered priceless to accept the diagnosis and see an exit for the impasse in which they found themselves. In addition, family and close friends were considered a very valuable source of support, in contrast to co-workers: given the long-term course of the condition, even if initially understanding of the situation, sustained support from co-workers commonly faded over time.

With regards to the school environment, many parents shared the view that the information they provided to the staff related to the VI of their children was not easily transferred from one year to the next, and they had to start the process of explaining the situation again. Besides, adapting the classroom layout and activities to the limitations of children with VI mainly rested on the goodwill of each of the teachers and, in those instances where this was absent, many incidents occurred, overtaxing both children and parents, inside and outside the classroom.

Other issues related to social support were the lack of proper coordination and collaboration between the administration, ONCE and the schools, the excessive bureaucracy involved in all administrative procedures, and the need to update assistive technology.

In summary, parents expressed that the sources of support they had to rely on did not always act as such.

**3.2.1.3. Boundary conditions.** The third subtheme included in the *obstacles race* main theme refers to the personal circumstances of the participants with an influence on the caregiving process and in their QoL. The impact of the VI of the children on family routines, the distribution of family roles, issues related to work/occupation, as well as the characteristics of the VI itself, may or may not act as added impediments.

Among the sample of children, there was a variety of ages, causes, severity and evolution of VI. However, the contribution of none of these variables was found to show a predominant influence in the *obstacles race*, according to the information collected from the interviews.

In terms of the distribution of roles, three of the participants noted in the sociodemographic survey that it was mainly the mother who adopted the caregiving role. The other participants stated that this responsibility was equally shared between father and mother. Interestingly, however, later discussion on this topic revealed that in those instances in which shared roles were initially reported, the main contribution to caregiving was still provided by the mother, although, when prompted to change their initial response, all participants chose to retain the shared roles option. Indeed, tasks mainly assumed by mothers were search for information, medical visits, administrative procedures, private tutoring, etc. This situation was reflected in the fact that those mothers not currently working, or having part-time or flexible hours jobs, chose these options to devote more time to caregiving tasks. On the contrary, all fathers worked full-time and none had reduced working hours or adopted a flexible work schedule.

Participants reported that the emotional involvement associated with the management of the situation was a source of stress in their relationship, but most of them avowed complementing each other well, offering mutual support, and felt the experience had made them grow and be more united as a couple.

### 3.2.2. Emotional impact subthemes

**3.2.2.1. Physical and psychological.** This subtheme describes the effect of caregiving on the physical and mental health of parents. In effect, all participants reported a high level of anxiety at least during the first years, and also deep sadness and fear. At the physical level, this had an impact on sleep and rest, muscle aches and constant fatigue.

[Participant 7] “My life collapsed. I was anticipating everything that would befall us.” [Participant 9] “Some years were horrific... I couldn't find my way out. I was crying all the time. I was overcome by the circumstances.”

Two cases progressed to clinical depression requiring psychotherapeutic and pharmacological treatment. Other ways in which the initial suffering was expressed were feelings of helplessness, lack of control, anguish, obsession, or recurring thoughts of guilt: guilt for not having noticed the signs before, for feeling responsible for the disease (“what have I done wrong?”, “he inherited it from me”), for paying less attention to their other children (brothers and sisters), for being unable to better organize time at work and care, etc. Some parents reported anger as their main feeling.

The lack of treatment and the unpredictability of the evolution of the condition was a constant source of anxiety, although participants disclosed that after a certain time physical symptoms and intensity of suffering decreased, without disappearing completely.

[Participant 2] “I'm better now... but it's something that's always there, you are always thinking about it.”

Other stressful elements were identified, related to day-to-day life: to be permanently alert to school issues, frustration with medical visits, lack of expected support, communication difficulties with health professionals and teachers, discomfort for being constantly challenged and the burden derived from the adopted roles.

**3.2.2.2. Locus of control.** This subtheme outlines the circumstances that lead to the exercise of internal control or external control. [Rotter \(1966\)](#) defines locus of control as the degree to which individuals perceive an outcome as being contingent on their own actions or those of external forces, existing along a continuum from a more internalized to a more externalized orientation. The purpose of this subtheme in our work consists in determining whether the attribution of an external locus of control is adopted when circumstances objectively do not depend on oneself, whereas internal locus of control attribution is triggered when one's actions may lead to a better adaptation to the situation. This subtheme is closely connected with the next, *coping skills*, and it may act as its trigger or damper.

Data revealed that the issue of attribution of external control over the disease was not trivial. Objectively, direct control of the disease is out of reach of parents and an internal control attribution may lead to frustration; conversely, an external locus of control may foster a relationship of trust with the health professional.

The intense and negative emotional reactions that all parents had experienced are common in such extreme circumstances. The ability to exert internal control over these responses is often very limited. In particular, parents referred to this limitation when describing their first reactions to the new situation; the regulation and dampening of the initial suffering took place as parents gained in ability to achieve internal control over their actions and feelings.

[Participant 7] “*I required therapeutic help and little by little I accepted it.*”

[Participant 8] “. *there is no other choice, either you move ahead, or you become stuck...*”.

In those occasions in which internal control of the caregiving process and its implications may be exerted, parents did so through the coping strategies described in the next section.

**3.2.2.3. Coping skills.** This subtheme refers to the cognitive and/or behavioral strategies that parents develop to deal with their caregiving roles. How do they face their lack of knowledge or the need for support? How do they modify boundary conditions? How do they manage their own emotions? In summary, what repertoire of coping strategies is available to them? Are they sufficient and adequate? How do these coping strategies influence *emotional impact* and, in turn, increase or alleviate the *weight on one's shoulders*?

Several coping strategies were disclosed. To combat negative feelings and perceived stress parents resorted to searching for information and support, practicing sport, or looking for some respite (as individuals or as a couple), among others. Several examples of cognitive reappraisal were also identified: stopping feeling responsible (and guilty) for the condition of the child, dispelling thoughts of threat or loss, and no longer focussing on the disability but on the person, by appreciating and trusting in the abilities of the children and fostering their autonomy.

[Participant 1] “... *I realized that I wasn't helping my son, or the family, or anyone, not even myself...*” [Participant 5] “*I reached the conclusion that it was positive to have time to myself.*”

A wide range of actions related to specific problems was compiled: altering boundary conditions (reducing or adopting flexible working hours, changing jobs, actively asking for the involvement of the rest of the family), applying for therapeutic support, increasing self-knowledge, getting involved in improving issues at school, conducting social awareness actions about VI, etc. Even though these strategies were commonly described as demanding tasks, all participants highlighted they would continue doing them.

It must be noted that some of the coping strategies parents used were unsuitable hence ineffective. These were mainly related to the initial period and based upon confrontation (e.g., with the health and education professionals) or avoidance (social isolation, passivity, focussing on the past). All of them were associated with stress and frustration.

**3.2.2.4. Positive expression.** The last subtheme of the *emotional impact* main theme underpins the change in perspective adopted by parents with time, as evidenced by positive expressions and acceptance. It may be assumed that this change was nurtured by a better balance regarding locus of control (internal or external) and by the implementation of adaptive coping strategies.

[Participant 4] “*We are now much calmer because we realize that with his visual acuity he can do much more than we thought two years ago.*” [Participant 6] “*For me the secret resides in thinking about what can be, not about what could have been if...*”.

It was observed that all participants focused mainly on the present, and admitted that as parents they may have magnified more their emotional impact on the condition of their children than the children themselves had. Indeed, they believed that their children lived their life with VI naturally.

## 4. Discussion

The thematic analysis of the transcribed interviews identified an overarching theme *the weigh on one's shoulders* and two main themes that modulate the perceived responsibility: *obstacles race* (including subthemes on knowledge of VI, support and boundary conditions) and *emotional impact* (physical and physiological aspects, locus of control, coping strategies and the ability for positive expression). Both sets of subthemes may influence one another: for instance, lacking the proper support in the school environment may lead to permanent alertness regarding the implementation of adjustments, time and money spent in private tutoring, poorer relationship with teachers, feelings of loss of control of the situation and increased stress and burden perception. On the contrary, acting pro-actively (e.g., meeting with other caregivers in a similar situation) may contribute to improving knowledge about VI, and to find necessary support and thus reduce *the weight on one's shoulders*.

The present findings revealed that even if VI itself was a factor leading to disability, additional aspects needed to be considered: the biopsychosocial model approach (Engel, 1977) allows contexts and behaviors to be considered as sources of disability, as they constrain the potential of children and challenge their QoL and that of their caregivers. In effect, the lack of knowledge and empathy regarding VI and its implications is a stressful experience for caregivers, with a negative impact on their perceived QoL. An insufficient knowledge, understanding and empathy reinforces the disability, leading to frustration if the adequate support is also absent.

This situation is commonly described within the field of rare diseases. For instance, Baumbusch et al. (2019) termed the experiences of 16 interviewed caregivers of children with rare diseases as being “alone in a crowd”, and those parents included in a study by Currie and Szabo (2018) noted that the joint impact of rare disease and disability significantly increased the burden of the family, using such self-explanatory terms as “navigators without a map” or “everything is under construction”. In this regard, an observational study exploring popular knowledge of the concepts “low vision” and “blindness” revealed that more than half of a sample of 188 participants

had never heard of “low vision” before and they did not associate “low vision” with systemic conditions such as type II diabetes (Lupón et al., 2021). Similarly, although almost all respondents were familiar with the term “blindness”, less than 30% of them were able to describe the sentence “blind people do not see anything” as false. Thus, the findings of that study are in agreement with the observations of the parents of children with VI interviewed in the present research.

All participants searched for information online by their own initiative, to improve their knowledge of the condition and to overcome the lack of information they felt was shared by health professionals. To improve the management of their time and their ability to find easily comprehensible, useful and reliable resources, all of them would have welcomed professional guided assistance, a shared finding with previous research employing similar methodologies (Baumbusch et al., 2019; Benedicta et al., 2020; Currie & Szabo, 2019; Jackson et al., 2007).

Participants expressed they expected to receive the explicit support of health and education professionals, and that of the administration, to ease their *obstacles race*. They also observed the lack of immediate attention protocols for families receiving a distressful diagnosis, and the overall lack of empathy. Previous research on caregiving children with VI (Jackel et al., 2010; Ek, 2000) or other chronic conditions also evidenced these findings (Courtney et al., 2018; Jackson et al., 2007; Ryerson Espino et al., 2018). A recent publication by Enoch et al. (2021) also describes the continuous need for informative support particularly addressed to their specific circumstances expressed by informal caregivers of individuals with VI.

In Catalonia, in particular, in mild, moderate (low vision) or unspecified VI, formal education support is provided only in exceptional occasions (e.g., tutoring of itinerant specialist teachers or assistive technology). Nevertheless, even with the assistance of itinerant specialist teachers, the engagement of the rest of the teachers and of the whole institution is required. Unfortunately, not only is this uncommon, but there is a lack of regulatory mechanisms to ensure it, and when VI is moderate or unspecified, involvement is often not regarded as urgent or critical. Therefore, it would seem that the school environment is not always enabling, but the opposite.

The severity or type of disability were not found to influence the *obstacles race*. Contradictory findings are evidenced in the literature in this regard. For instance, Rahi et al. (2005) noted that higher satisfaction of parents was associated with less severe disabilities and absence of comorbidities. In contrast, Sola-Carmona et al. (2016) described higher self-esteem in parents of blind children than in those with less severe VI.

Caregiving was found to be a role predominantly assigned to mothers, either explicitly or tacitly, even when this resulted in the need to work flexible hours or reduce working hours, with the corresponding implications for job promotion, salary and self-realization. Nevertheless, some mothers opted to formally express equally shared roles, which may raise the question of whether some mothers feel uncomfortable to openly admitting that roles are not balanced, or that they simply accept and adopt a role traditionally assigned to them. In effect, qualitative research by Jiménez and Moya (2018) describes both false consensus and interpretative bias related to the “feminine” nature of caregiving: there is a consensus that caregiving is ‘something natural’ and a ‘moral obligation’ of women and, even if male involvement is insignificant, it is biasedly interpreted as of similar magnitude.

The present findings revealed the emotional impact of caregiving a child with VI. Both before and after the initial diagnosis, participants reported great suffering and fear because of the uncertainty of the prognosis, lack of control, ignorance, and collapse of expectations, among others. Other participants noted feelings of guilt and anger. These feelings, emotions and thoughts were also documented in studies by Cardinali et al. (2019) and Gómez-Zúñiga et al. (2019) based on interviews of parents of children with rare diseases in Italy and Spain.

Given the open format of the interviews, parents were offered the opportunity to describe the experienced changes in physical and psychological impact with time: at the time, most of them were feeling better, although the state of alertness and a certain level of grief never disappears. Time allowed them a better understanding of both the condition causing VI and of VI itself, and everything that it entails, as well as a perception of greater internal control in the caregiving process arising from a wide variety of coping strategies. All participants reached the conclusion that, although the visual condition of their children is irreversible, their own discouragement is not, and they are pro-active in spite of the adversities.

Hence, the present investigation leads us to consider that informal caregiving of children with VI has a negative influence on all domains of QoL of their parents, with a particular impact on the psychological and environment domains described by the WHOQoL-BREF questionnaire. The negative impact on the physical health domain predominantly occurs before a diagnosis of the condition is reached, and some time afterwards. Caregiving leads to a permanent level of stress, with an initial very intense emotional response and recurrence of negative feelings associated with the actual diagnosis (rare disease, chronicity, uncertain prognosis, etc.) which tend to decrease with time. However, a certain level of basal stress remains, fueled by feelings of anguish and sadness caused by the perception of lack of knowledge and empathy of health professionals, the perceived gap between the expectations and need for support of caregivers and the actual formal support provided to them, and the absence of control over and treatment of the condition causing VI. In the social relationships’ domain, unbalanced gender roles were evidenced, but participants considered this a minor concern. Finally, the environment domain is mainly associated with job related opportunities, training, self-realization, and management of leisure time, with a negative impact on the other domains if caregivers do not adopt the corresponding adjustments. This domain is also influenced by the insufficient care and support towards children with VI and families provided by health professionals and public administrations.

#### 4.1. Strengths and limitations

As far as a previous literature research revealed, no similar qualitative analysis of QoL of caregivers of children with VI has been conducted in Catalonia (or the rest of Spain). The outcome of the thematic analysis of the transcribed interviews according to the guidelines of Braun and Clarke (2006) revealed an overarching theme, two main themes and several subthemes, which were in turn

interpreted within the framework of QoL literature and of the domains of the WHOQoL-BREF questionnaire.

Given the low prevalence of VI in children, sampling was intentional and some intrinsic limitations were unavoidable. For instance, the sample was reduced (9 participants), with more mothers ( $n = 6$ ) than fathers ( $n = 3$ ), all of them from nuclear families, with at least secondary education and holding full or part-time jobs. It would be interesting to investigate whether the same themes and subthemes would arise in a sample including participants with lower educational levels, unemployed, or from different family structures.

## 5. Conclusion

A qualitative approach on the QoL of caregivers of children with VI evidenced the influence caregiving has on all QoL domains of parents. The analysis revealed a core theme to describe the persistent psychological distress that parents experience due to multiple factors, both intrinsic and extrinsic.

Although this qualitative analysis was based on children with VI in Catalonia, it may be assumed that similar findings would be revealed in the QoL of caregivers of children with other rare conditions in Catalonia in particular, and in Spain. Further research may explore whether these findings are similar in other European countries as it may be hypothesized that, sharing similar difficulties in front of rare diseases associated with VI, other European parents and caregivers may experience comparable problems. Health and education professionals and public administrations are encouraged to develop strategies to improve the support and information provided to these families at each stage of their challenging situation.

## Ethical approval

All procedures involving participants were conducted in accordance with the ethical standards of the University Hospital Mutua de Terrassa, Act 02/2018 (Catalonia, Spain), and followed the Declaration of Helsinki. Informed consent was obtained from all participants after the objectives of the study were explained to them, and the corresponding data protection policies were implemented.

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## CRedit authorship contribution statement

**Marta Lupón:** Conceptualization, Methodology, Formal analysis, Investigation, Resources, Writing - original draft, Writing - review & editing, Visualization. **Manuel Armayones:** Conceptualization, Writing - review & editing, Visualisation, Supervision. **Genís Cardona:** Conceptualization, Writing - review & editing, Visualisation, Supervision.

## Declaration of Competing Interest

None.

## Data availability

Data will be made available on request.

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## References

- Al Qadire, M., Aloush, S., Alkhalileh, M., Qandeel, H., & Al-Sabbah, A. (2020). Burden among parents of children with cancer in Jordan: prevalence and predictors. *Cancer Nursing*, 43(5), 396–401.
- Baumbusch, J., Mayer, S., & Sloan-Yip, I. (2019). Alone in a crowd? Parents of children with rare diseases' experiences of navigating the healthcare system. *Journal of Genetic Counseling*, 28(1), 80–90.
- Benedicta, B., Caldwell, P. H., & Scott, K. M. (2020). How parents use, search for and appraise online health information on their child's medical condition: A pilot study. *Journal of Paediatrics and Child Health*, 56(2), 252–258.
- Bezabih, L., Abebe, T. W., & Fite, R. O. (2017). Prevalence and factors associated with childhood visual impairment in Ethiopia. *Clinical Ophthalmology*, 11, 1941–1948.
- Braun, V., & Clarke, V. (2006). Using thematic analysis in Psychology. *Qualitative Research in Psychology*, 3(2), 77–101.
- Cardinali, P., Migliorini, L., & Rania, N. (2019). The caregiving experiences of fathers and mothers of children with rare diseases in Italy: Challenges and social support perceptions. *Frontiers in Psychology*, 10, 1780.
- Commissariat, P. V., Harrington, K. R., Whitehouse, A. L., Miller, K. M., Hilliard, M. E., Van Name, M., DeSalvo, D. J., Tamborlane, W. V., Anderson, B. J., DiMeglio, L. A., & Laffel, L. M. (2020). I'm essentially his pancreas": Parent perceptions of diabetes burden and opportunities to reduce burden in the care of children <8 years old with type 1 diabetes. *Pediatric Diabetes*, 21(2), 377–383.



- Courtney, E., Kiernan, G., Guerin, S., Ryan, K., & McQuillan, R. (2018). Mothers' perspectives of the experience and impact of caring for their child with a life-limiting neurodevelopmental disability. *Child: Care, Health and Development*, 44(5), 704–710.
- Currie, G., & Szabo, J. (2018). 'It is like a jungle gym, and everything is under construction': The parent's perspective of caring for a child with a rare disease. *Child: Care, Health and Development*, 45(1), 95–103.
- Currie, G., & Szabo, J. (2019). 'It would be much easier if we were just quiet and disappeared': Parents silenced in the experience of caring for children with rare diseases. *Health Expectations: An International Journal of Public Participation in Health Care and Health Policy*, 22(6), 1251–1259.
- Ek, U. (2000). Emotional reactions in parents and children after diagnosis and treatment of a malignant tumour in the eye. *Child: Care, Health and Development*, 26(5), 415–428.
- Engel, G. (1977). The need for a new medical model: A challenge for Biomedicine. *Science*, 196(4286), 129–136.
- Enoch, J., Dickinson, C., & Subramanian, A. (2021). What support is beneficial for caregivers of individuals with vision impairment? *Clinical and Experimental Optometry*, 104(4), 455–470.
- Generalitat de Catalunya, & Departament d'Educació [Government of Catalonia, Education Department]. (2021). Estadística de l'ensenyament [Schooling Statistics]. Retrieved from: <https://educacio.gencat.cat/ca/departament/estadistiques/estadistiques-ensenyament/cursos-anteriors/curs-2020-2021/> Accessed July 2022.
- Gómez-Zúñiga, B., Pulido Moyano, R., Pousada Fernández, M., García Oliva, A., & Armayones Ruiz, M. (2019). The experience of parents of children with rare diseases when communicating with healthcare professionals: Towards an integrative theory of trust. *Orphanet Journal of Rare Diseases*, 14(1), 159.
- Gyawali, R., Bhayal, B. K., Adhikary, R., Shrestha, A., & Prasad Sah, R. (2017). Retrospective data on causes of childhood vision impairment in Eritrea. *BMC Ophthalmology*, 209(17).
- Hatton, D. D., Ivy, S. E., & Boyer, C. (2013). Severe visual impairments in infants and toddlers in the United States. *Journal of Visual Impairment & Blindness*, 107(5), 325–336.
- Haugen, O. H., Bredrup, C., & Rødahl, E. (2016). Visual impairment in children and adolescents in Norway. *Tidsskrift for den Norske lægeforen*, 136(11), 996–1000.
- IMRSERO, Instituto de Mayores y Servicios Sociales [Institute for the Elderly and Social Services]. (2022). Base estatal de datos de personas con valoración del grado de discapacidad. Informe a 31/12/2020 [State database of people with disability. Report on 12/31/2020]. Retrieved from: [https://www.imsero.es/InterPresent2/groups/imsero/documents/binario/bdepcd\\_2020.pdf](https://www.imsero.es/InterPresent2/groups/imsero/documents/binario/bdepcd_2020.pdf) Accessed September 2022.
- Jackel, B., Wilson, M., & Hartmann, E. (2010). A survey of parents of children with cortical or cerebral visual impairment. *Journal of Visual Impairment & Blindness*, 104(10), 613–623.
- Jackson, R., Baird, W., Davis-Reynolds, L., Smith, C., Blackburn, S., & Allsebrook, J. (2007). Qualitative analysis of parents' information needs and psychosocial experiences when supporting children with health care needs. *Health Information and Libraries Journal*, 25(1), 31–37.
- Jiménez, I., & Moya, M. (2018). The family caregiver: The naturalized sense of obligation in women to be caregivers. *Enfermería Global*, 49(1), 434–447.
- Lupón, M., Armayones, M., & Cardona, G. (2018). Quality of life among parents of children with visual impairment: A literature review. *Research in Developmental Disabilities*, 83, 120–131.
- Lupón, M., Cardona, G., & Armayones, M. (2021). Public knowledge of low vision and blindness, and readability of on-topic online information. *Journal of Optometry*, 14(3), 240–246.
- Picardi, A., Gigantesco, A., Tarolla, E., Stoppioni, V., Cerbo, R., Cremonte, M., Alessandri, G., Lega, I., & Nardocci, F. (2018). Parental burden and its correlates in families of children with Autism Spectrum Disorder: A multicentre study with two comparison groups. *Clinical Practice and Epidemiology in Mental Health*, 14, 143–176.
- Rahi, J. S., & Cable, N. (2003). Severe visual impairment and blindness in children in the UK. *The Lancet*, 362(9393), 1359–1365.
- Rahi, J. S., Manaras, I., Tuomainen, H., & Lewando-Hundt, G. (2005). Health services experiences of parents of recently diagnosed visually impaired children. *The British Journal of Ophthalmology*, 89(2), 213–218.
- Resnikoff, S., Pascolini, D., Etya'ale, D., Kocur, I., Pararajasegaram, R., Pokharel, G., & Mariotti, S. P. (2004). Global data on visual impairment in the year 2002. *Bulletin of the World Health Organization*, 82(11), 844–851.
- Rotter, J. B. (1966). Generalized expectancies for internal versus external control of reinforcement. *Psychological Monographs: General and Applied*, 80(1), 1–28.
- Ryerson Espino, S. L., Kelly, E. H., Rivelli, A., Zebracki, K., & Vogel, L. C. (2018). It is a marathon rather than a sprint: an initial exploration of unmet needs and support preferences of caregivers of children with SCI. *Spinal Cord*, 56(3), 284–294.
- Sola-Carmona, J. J., López-Liria, R., Padilla-Góngora, D., Daza, M. T., Aguilar-Parra, J. M., & Salido-Campos, M. A. (2016). Factors associated with the anxiety, subjective psychological well-being and self-esteem of parents of blind children. *PLoS ONE*, 11(9), Article e0162294.
- Solebo, A. L., & Rahi, J. (2014). Epidemiology, aetiology and management of visual impairment in children. *Archives of Disease in Childhood*, 99(4), 375–379.
- Solebo, A. L., Teoh, L., & Rahi, J. (2017). Epidemiology of blindness in children. *Archives of Disease in Childhood*, 102(9), 853–857.
- WHO, World Health Organization, (1998). Division of Health Promotion, Education, and Communication. Health promotion glossary. Retrieved from: <https://apps.who.int/iris/handle/10665/64546> Accessed February 2022.
- WHO, World Health Organization, (2012). The World Health Organization Quality of Life (WHOQOL). Retrieved from: <https://www.who.int/publications/i/item/WHO-HIS-HSI-Rev.2012.03> Accessed February 2022.
- WHO, World Health Organization, (2020). ICD-11 for Mortality and Morbidity Statistics: Retrieved from: <https://icd.who.int/browse11/m/en#/http%3a%2f%2fid.who.int%2fid%2fentify%2f1103667651> Accessed March 2022.
- WHO, World Health Organization, (2021). Blindness and vision impairment. Retrieved from: <https://www.who.int/news-room/fact-sheets/detail/blindness-and-visual-impairment> Accessed June 2022.
- Zaidman-Zait, A., Most, T., Tarrasch, R., Haddad-Eid, E., & Brand, D. (2016). The impact of childhood hearing loss on the family: Mothers' and Fathers' stress and coping resources. *The Journal of Deaf Studies and Deaf Education*, 21(1), 23–33.