

Parent-child discrepancy in the assessment of health-related quality of life using the EQ-5D-Y questionnaire

Miguel Á. Pérez Sousa, M.D.,^a Pedro R. Olivares Sánchez-Toledo, M.D.,^b and Professor Narcis Gusi Fuerte^a

ABSTRACT

Introduction. The assessment of health-related quality of life (HRQoL) serves to detect changes over time in patients' health status and allows to do a cost-effectiveness analysis of treatments. When children with special health features cannot perform a self-assessment, it is possible to assess their HRQoL through their parents or caregivers. To date, the discrepancy in the assessment of HRQoL using the EQ-5D-Y questionnaire among children with cerebral palsy (CP) and their parents has not been analyzed. The objective of this study was to analyze the level of agreement in the HRQoL assessment using the EQ-5D-Y questionnaire and its proxy version among children with CP and their parents or caregivers.

Population and methods. Children and adolescents with CP, and their parents, from a special education school in the region of Extremadura (Spain) participated in the study. The EQ-5D-Y questionnaire was used for children and the EQ-5D-Y proxy version, for parents. Interviews were conducted in the first quarter of 2015. The level of agreement in the responses was analyzed using the Cohen's kappa coefficient for the five domains of the EQ-5D-Y and the intraclass correlation coefficient for the visual analogue scale.

Results. Sixty-two children with CP and mild and/or moderate functional capacity impairment, and their parents, participated in the study. The level of agreement was poor in the HRQoL assessment between children and parents in all the questionnaire domains (<0.20) and fair or poor (<0.60) in the visual analogue scale.

Conclusions. A high level of parent-child disagreement was observed in the HRQoL assessment in the population with CP using the EQ-5D-Y questionnaire.

Key words: Health-related quality of life, proxy, agreement, cerebral palsy.

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INTRODUCTION

The assessment of health-related quality of life (HRQoL), understood as a multidisciplinary term that encompasses medical, physiological, and biochemical outcomes,¹ has spread over the past decade and resulted in a wider availability of generic questionnaires such as the PedsQL,² Kidscreen,³ and EuroQol 5D.⁴ Although it is true that these questionnaires for the self-assessment of HRQoL have been validated among different populations with special health features, there are certain populations who, given their distinctive health or sociocultural characteristics, are unable to perform a self-assessment; this leads to the emergence of the external observer role, who is in direct relation with the patient. For this reason, different versions of HRQoL assessments have been developed that serve to rate it by means of a proxy, usually parents, legal tutors or health care agents.

Over the past years, a proxy version has been developed of the main HRQoL assessment instruments, such as the EQ-5D-Y, the Kidscreen, and the PedsQL, and their reliability and validity have been demonstrated.⁵⁻⁹

Given that HRQoL in children and adolescents is the object of our study, there is a wide range of possibilities to assess it because proxy questionnaires can be applied to children and adolescents who have limiting conditions and also to those with reading comprehension difficulties or who have a low socioeconomic level. In this regard, parents are fundamental to assess their children's HRQoL¹⁰ and make critical contributions to treatment-

- a. School of Sports Science, Universidad de Extremadura.
- b. Research Institute of Physical Activity and Health, Universidad Autónoma de Chile, Talca, Chile. olivares.pedro@gmail.com.

E-mail address:

Miguel Á. Pérez Sousa, M.D.: maperezsousa@unex.es.

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related decisions that will be implemented by physicians and specialists,^{11,12} and to detect changes over time in their children's HRQoL.¹³ However, the assessment made by a proxy may never replace the child/patient who could perform it correctly because they provide a more accurate assessment of psychosocial domains.^{14,15}

In different studies, a higher or lower level of agreement or disagreement has been described between children/patients and their proxies both in the healthy population and in that with special health features.¹⁴⁻¹⁹ In relation to children with motor disorders, such as cerebral palsy (CP), a high level of disagreement has been observed between children and parents, and their children's HRQoL has been assessed as poor,¹⁷ as is the case of children with muscular dystrophy.²⁰ Therefore, there is a lot of ambiguity in relation to the level of agreement between the proxies and children in the healthy population and that with special health features because of the difficulty entailed by the population itself or the extent of questionnaires used.

In this line, the proxy version of the EQ-5D-Y questionnaire, adapted and validated into Spanish,⁶ has the same characteristics than the proxy version for adults and elderly people. It is short, easily administered, and provides results on different health domains and an index value that can be used to assess health status. It is also useful for the economic assessment analysis of health care services. However, the level of agreement/disagreement between subjects/patients and their proxies has not been studied among children with CP.

The objective of this study was to analyze the level of agreement between children with CP and the healthy population, and also among their parents, using the EQ-5D-Y questionnaire and its proxy version. The hypothesis proposed here was that there was a high level of disagreement in the HRQoL assessment between parents and their children with CP.

POPULATION AND METHODS

Participants

To select the sample, three special education schools from the Extremadura community were contacted. The research protocol was first presented to the school's administration and the parent's council. One of the special education schools for people with CP agreed to participate in the study and signed the informed consent.

The study was conducted in the first quarter

of 2015. Children between 6 and 17 years old with CP from a specialized facility, classified based on their functional capacity as grade 1 CP (without activity limitations) or as grade 2 CP (mild or moderate activity limitations), as well as their parents, were included in the study. Both children and parents were asked to sign the informed consent as a requirement for participation.

Children completed the self-report version and parents, the proxy version of the EQ-5D-Y questionnaire.

The following were the inclusion criteria for participants:

1. Having grade 1 CP (without activity limitations) or grade 2 CP (mild or moderate activity limitations), as classified by the health care team/therapist from the school based on functional capacity.
2. Having an adequate reading comprehension level to understand items and answer questions.
3. Being present on the day of the test.
4. Signing the informed consent (subjects and parents or legal tutors).

These inclusion criteria were managed by the school's therapist team.

Interviews were conducted in the classrooms prepared to this end by a technician experienced in this type of tests. The questionnaire, which lasted approximately 8 minutes, was administered by means of individual interviews.

An appointment was scheduled with the parents at the corresponding school, and interviews were conducted with mothers in one classroom and fathers in a separate classroom. The study was approved by the Bioethics Committee of Universidad de Extremadura and conducted in accordance with the International Ethical Guidelines for Biomedical Research Involving Human Subjects, established in Geneva.

Instruments

EQ-5D-Y questionnaire and its proxy version: The Spanish version of the EQ-5D-Y questionnaire²¹ and its proxy version⁶ include a descriptive section made up of five domains with three answer levels ("no problems," "some problems," and "a lot of problems"). The questionnaire domains are mobility, self-care, usual activities, pain and discomfort, and anxiety and depression. It also includes a visual analogue scale (VAS) for the subject to perform an overall assessment of his/her health status in a scale from

0 to 100, where 0 means the worst health status and 100, the best health status.

The proxy version, which is completed by parents in relation to their children’s HRQoL, has the same characteristics of the self-report version.

This is a questionnaire to assess HRQoL in relation to five key aspects of individuals’ quality of life: ability to walk about, activities related to self-care (washing or dressing themselves), daily living activities (going to school, sports or doing things with family and friends), pain or discomfort, and lastly, anxiety or depression. Finally, the questionnaire describes the frequency of the problems reported by the subject or, in the case of the proxy version, by an external observer who is in direct relation with the child/patient.

The EQ-5D-Y questionnaire and its proxy version were tested previously by the research team evaluators using the Evaluating the Measurement of Patient-Reported Outcomes (EMPRO) tool; both versions obtained a high score in the areas of reliability, validity, interpretability, burden, alternative modes of administration, and cross-cultural and linguistic adaptations. In addition, the questionnaire has been extensively used worldwide in different populations with special health features,²²⁻²⁵ and its metric properties have been described in its Spanish version for Spain,⁶ as well as its comprehensibility and cultural adaptation in the general population.⁷ In this regard, a pilot test was done in advance to assess the questionnaire’s comprehensibility using query and paraphrasing methods in the target population, and it demonstrated an optimal comprehensibility.

Questions on the frequency of health problems in the past 6 months were also included, such as

headache, stomach pain, back pain, low mood, irritability, nervousness or trouble sleeping, together with questions on sociodemographic aspects, such as sex, age, country of origin or language.

Analysis

The sample size was estimated for a significance level of 0.05 and a confidence level of 90%.

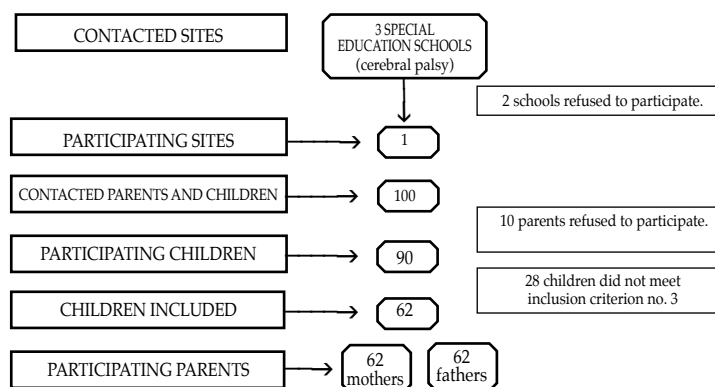
The statistical analysis was performed with the SPSS 21.0 software. To establish data normality and distribution, mean values, standard deviation, and frequencies were used. The level of agreement in the responses to the five domains of the EQ-5D-Y questionnaire and its proxy version was analyzed using Cohen’s kappa coefficient in accordance with Landis and Koch’s criteria,²⁶ and the following levels of agreement were established: null: <0; poor: 0-0.20; weak: 0.21-0.40; moderate: 0.41-0.60; good: 0.61-0.80; and very good: 0.81-1.00, together with an intraclass correlation coefficient (ICC) for the VAS. ICC values corresponded to null agreement: <0.20; poor agreement: 0.21-0.40; fair agreement: 0.41-0.59; good agreement: 0.60-0.74; and excellent agreement: 0.75-1.0. A descriptive, cross-sectional design was used in this research.

RESULTS

Participants

The flowchart of participants (*Figure 1*) shows that 90 children and their parents agreed to participate in the study. Some of them did not attend the school on the day of the test, so a total of 62 children with CP, and their parents, took part in the study. *Table 1* describes the sample characteristics and shows a mild difference in

FIGURE 1. Flowchart of participants



terms of sex distribution and a predominance of middle socioeconomic level in the overall sample.

TABLE 1. Sample characteristics. N= 62 children with cerebral palsy and their parents

	(n = 62)
Age (years), M (SD)	
Children	10.09 (SD: 2.31)
Fathers	41.2 (SD: 7.8)
Mothers	40.7 (SD: 5.8)
Sex, n (%)	
Boys	35 (56.4)
Girls	27 (43.6)
Country of origin	
Spain	62 (100.0)
Other	0 (0.0)
Socioeconomic level, n (%)	
Low	12 (19.4)
Middle	38 (61.2)
High	12 (19.4)
Functional classification of CP	
Grade 1	41 (66.1)
Grade 2	21 (33.9)

M: mean; SD: standard deviation; CP: cerebral palsy.

Frequency of problems

Table 2 shows the prevalence of problems reported by children and their parents. There were no missing values.

In general, it can be seen that fathers reported a lower frequency of problems in the EQ-5D-Y domains, especially in relation to self-care, usual activities, and anxiety or depression. In addition, a mild overestimation of health in the assessment made by fathers was observed based on the VAS. However, mothers report a higher number of problems than fathers and make a lower assessment of their children's health status.

Level of agreement

Table 3 describes the level of agreement between children and mothers, children and fathers, and mothers and fathers in the population with CP. A low level of agreement was observed across all comparisons and in most domains. It is very worth noting that the level of agreement whenever fathers are involved in assessing HRQoL is poor. In addition, although not high, the level of agreement between mothers and children is closer in the mobility and usual

TABLE 2. Responses to the domains of the EQ-5D-Y questionnaire and its proxy version. N: 62

EQ-5D-Y domains	Cerebral palsy					
	Children (n= 62)		Mothers (n= 62)		Fathers (n= 62)	
	%	n	%	n	%	n
Mobility						
I have no problems walking about.	25.8	16	27.4	17	33.9	21
I have some problems walking about.	58.1	36	45.2	28	54.8	34
I have a lot of problems walking about.	16.1	10	27.4	17	11.3	7
Looking after myself						
I have no problems washing or dressing myself.	30.6	19	54.8	34	61.3	38
I have some problems washing or dressing myself.	53.2	33	38.7	24	35.5	22
I have a lot of problems washing or dressing myself..	16.1	10	6.5	4	3.2	2
Doing usual activities						
I have no problems doing my usual activities..	53.2	33	58.1	36	74.2	46
I have some problems doing my usual activities.	30.6	19	35.5	22	25.8	16
I have a lot of problems doing my usual activities.	16.1	10	6.5	4	0.0	0
Having pain or discomfort						
I have no pain or discomfort.	45.2	28	58.1	36	69.4	43
I have some pain or discomfort.	46.8	29	29.0	18	22.6	14
I have a lot of pain or discomfort.	8.1	5	12.9	8	8.1	5
Feeling worried, sad or unhappy						
I am not worried, sad or unhappy.	53.2	33	62.9	39	83.9	52
I am a bit worried, sad or unhappy.	41.9	26	33.9	21	14.5	9
I am very worried, sad or unhappy.	4.8	3	3.2	2	1.6	1
VAS	70.4	12.6	76.2	14.5	80.8	14.3

VAS: visual analogue scale; SD: standard deviation.

Values were expressed as percentage, except for VAS, which was expressed as median (SD).

activities, with an acceptable (0.713) and moderate (0.436) agreement, respectively.

DISCUSSION

According to the current bibliography, this is the first study on the level of agreement between fathers and children, mothers and children, and fathers and mothers using the EQ-5D-Y questionnaire to assess HRQoL administered to children and adolescents with CP. The following were the three main findings of this research. First of all, fathers report fewer problems than mothers and children themselves in all domains. Secondly, there is a high level of disagreement between fathers and mothers in all domains. Lastly, it is worth noting that there is an absolute discrepancy between fathers and children and mothers and children.

A low level of agreement has been described between fathers and children in studies conducted using the Kidscreen questionnaire for children with CP,¹⁷ as observed with the PedsQL questionnaire, which showed little agreement in the emotional domain.²⁷ These findings support our hypothesis that there is a low level of agreement in psychosocial and physical functions, and those results are similar to the ones observed in our study. Therefore, parents of children with CP report a low HRQoL in general and greater psychological problems than children themselves, which was a tendency observed in previous studies, as described here.

This may be explained by the tendency of children to emphasize the positive aspects of their disease or their ability to adapt to changes.²⁸ This way, parents may be more reliable to assess which of their children capabilities are more affected because they may offer information on

the difficulties that children themselves attempt to hide.²⁷

Besides, a high level of disagreement was observed between children’s self-assessment and that of fathers, which is contradictory to what previous studies suggested that fathers served better as proxies than mothers.²⁹ However, that study was conducted among children with asthma, whose capabilities are not as limited as in our target population.

This was also observed in our study, in the HRQoL assessment made by fathers, whose level of agreement is not as high as that of mothers, and this may be explained by the fact that they spend less time in their children’s upbringing. Although the level of agreement between mothers and children was low in our study, this dyad showed a higher agreement than the fathers-children dyad. HRQoL assessment is mostly in the hands of mothers,^{30,31} therefore, it is believed that basing an assessment only on the mothers may somehow distort results. Although mothers are usually responsible for their children’s care, there are certain aspects that should be taken into consideration to understand the assessment of their children’s HRQoL, for example, the extent to which they are affected, which will establish a tendency towards agreement or disagreement. This situation may make mothers act in a more protective manner or, on the contrary, and as seen before, given the stress caused by looking after their children, it may make them underestimate their health.

This study has certain limitations. Particularly, the small size of the target population with CP, which hinders the possibility of obtaining evidence. Also, based on inclusion criteria, only patients with grade 1 and 2 CP were included.

TABLE 3. Level of agreement between parents and children with cerebral palsy

	Child-father (n= 62)			Child-mother (n= 62)			Father-mother (n= 62)		
	Kappa	P value	Level of agreement	Kappa	P value	Level of agreement	Kappa	P value	Level of agreement
Mobility	0.042	0.653	Poor	0.713	0.000*	Good	0.054	0.545	Poor
Self-care	0.044	0.622	Poor	0.057	0.536	Poor	0.046	0.680	Poor
Usual activities	0.019	0.841	Poor	0.436	0.000*	Moderate	0.182	0.096	Poor
Pain or discomfort	0.067	0.469	Poor	0.128	0.183	Poor	0.010	0.918	Poor
Anxiety or depression	0.016	0.854	Poor	0.165	0.140	Poor	0.030	0.765	Poor
VAS (ICC)	0.581	0.962	Fair	0.389	0.029	Poor	0.006	0.510	Null

* p < 0.05.

The levels of agreement were expressed as Cohen’s kappa coefficient for the EQ-5D-Y domains and as intraclass correlation coefficient (ICC) for the visual analogue scale (VAS).

Another weakness of this study was the use of only one HRQoL questionnaire.

Future lines: to increase the sample size to draw more solid conclusions, administer a larger set of questionnaires to collect more information and thus compare their implementation, even across different populations.

CONCLUSIONS

The main conclusion of this research is that there is a high level of parent-child disagreement in HRQoL assessment in the population with CP using the EQ-5D-Y questionnaire. ■

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