

2023

**Assessing the Applicability and Appropriateness of Quality of Life
measurement Instruments for Children with Cerebral Palsy in Low- and
Middle-Income Countries: A Qualitative Study**

Bobbi van Kesteren
Switzerland

Master of Science in International Health
KIT (Royal Tropical Institute)
Vrije Universiteit Amsterdam (VU)



KIT Royal
Tropical
Institute

Assessing the Applicability and Appropriateness of Quality of Life measurement Instruments for Children with Cerebral Palsy in Low- and Middle-Income Countries: A Qualitative Study

A thesis submitted in partial fulfillment of the requirement for the degree of Master of Science in International Health

by

Bobbi van Kesteren.

Declaration:

Where other people's work has been used (from either a printed or virtual source, or any other source), this has been carefully acknowledged and referenced in accordance with academic requirements. The thesis "Assessing the Applicability and Appropriateness of Quality of Life measurement Instruments for Children with Cerebral Palsy in Low- and Middle-Income Countries: A Qualitative Study" is my own work.



Signature:

Master of Science in International Health (MIH)
2 December – 9 Augustus 2023
KIT (Royal Tropical Institute)/Vrije Universiteit Amsterdam
Amsterdam, The Netherlands

Augustus 2023

Organized by:
KIT (Royal Tropical Institute)
Amsterdam, The Netherlands

In cooperation with:

Vrije Universiteit Amsterdam (VU)
Amsterdam, The Netherlands

Abstract

Introduction:

Cerebral palsy is one of the major causes of childhood disability, affecting children's quality of life world-wide. In high-income countries, children with cerebral palsy are assisted by a specialized multidisciplinary team of healthcare and rehabilitation professionals. Low- and middle-income countries rely on other more basic sources of care, particularly family-centered care and community based rehabilitation. To evaluate the effect of the applied interventions, quality of life can be used as an outcome measure. Quality of life can be measured with generic measurement tools as well as with disease-specific measurement tools. Most of the instruments developed for measuring quality of life in children with cerebral palsy, are developed in high-income countries. This could limit their applicability and appropriateness in low- and middle-income countries, which could hinder efforts to improve care and rehabilitation. Therefore, the aim of this study was to investigate the extent to which existing quality of life measurement instruments are applicable and appropriate for evaluating quality of life in children with cerebral palsy residing in low- and middle-income countries.

Methodology:

An extensive literature search was performed followed by a primary qualitative analysis. Thereafter, semi-structured in-depth interviews with 11 field experts were undertaken to gain insight into their perceptions on measuring quality of life for children with cerebral palsy in low- and middle-income countries. The data was then systematically coded and grouped into categories through a thematic analysis.

Results:

Four themes emerged from the interviews held with experts namely: 1) Conceptualizing QoL, 2) Key QoL domains, 3) Existing QoL measurement tools and 4) Experts insights on tool usage for CP children in LMICs. Experts assert that the interpretation of quality of life varies between children with cerebral palsy residing in high-income countries and those in low- and middle-income countries. Quality of life key domains that were identified by experts were Beliefs & Stigma, Family, Resources & Access, Education and Food & Malnutrition.

Discussion & conclusion

The results found in this study reveal both similarities and discrepancies when compared to existing literature. Evidently, the cerebral palsy quality of life questionnaire for children stands out as the most robust measurement tool for evaluating the quality of life in children with cerebral palsy in low- and middle-income countries. Nevertheless, its utilization warrants caution and meticulousness, considering cultural and contextual nuances. Notably, this measurement instrument lacks the inclusion of two important aspects that experts highlighted: beliefs & stigma, as well as the inclusion of feeding strategies. In summation, it proves to be the strongest quality of life measurement tool at present. However, forthcoming research should prioritize the development of a more culturally appropriate quality of life measurement instrument for children with cerebral palsy in low- and middle-income countries.

Keywords: Cerebral palsy, children, quality of life, appropriateness, applicability, measurement instruments, qualitative research and low- and middle-income countries.

Word count

11.570

Table of content

Abstract.....	ii
Table of content	iii
List of Tables.....	v
List of Figures.....	v
Abbreviations	vi
Glossary	vii
Acknowledgements	viii
1) Introduction.....	ix
2) Background.....	1
3) Problem statement, justification and objectives.....	3
3.1 Problem statement	3
3.1.1 Measurement tools	3
3.1.2 Culture and context	4
3.2 Justification	4
3.3 Objectives.....	5
4) Methodology	6
4.1 Study design.....	6
4.2 Participants.....	6
4.3 Data collection	6
4.3.1 Literature review.....	6
4.3.2 Semi-structured in-depth interviews	7
4.4 Data analysis.....	7
4.4.1 Analysis of literature	7
4.4.2 Analysis of semi-structured in-depth interviews	7
4.4.3 Rigour.....	8
4.5 Ethics and integrity.....	8
5) Results	9
5.1 Characteristics experts	9
5.2 Conceptualizing QoL	10
5.3 Key QoL domains.....	11
5.3.1 Beliefs & Stigma	11

5.3.2 Family.....	11
5.3.3 Resources & Access.....	12
5.3.4 Education.....	13
5.3.5 Food & Malnutrition.....	13
5.4 Existing tools.....	16
5.5 Experts insights on tool usage for CP children.....	22
6) Discussion.....	25
6.1 Domains.....	25
6.2 CP-QoL CHILD.....	26
6.3 Limitations and strengths.....	29
7) Conclusion and recommendations.....	31
8) References.....	33
9) Appendix.....	38
Appendix 1: Invitation email.....	38
Appendix 2: Informed Consent.....	39
Appendix 3: Topic guide.....	41
Appendix 4: Code Tree.....	43
Appendix 5: Ethical Clearance.....	57

List of Tables

Table 1: Overview experts details	9
Table 2: General features measurement tools	16
Table 3: Overview +/- existing measurement tools, literature	18
Table 4: Overview +/- existing measurement tools, experts	21

List of Figures

Figure 1: Illustration of 5 QoL domains	13
Figure 2: Data stratification QoL domains by experts	15
Figure 3: Scoring CP CHILD, CP-QoL CHILD, PedsQL, DISABKIDS	20

Abbreviations

AI	Artificial Intelligence
CBR	Community Based Rehabilitation
CF	Cystic Fibrosis
CHQ	Child Health Questionnaire
CP	Cerebral Palsy
CPCHILD	Caregiver Priorities and Child Health Index of Life with Disabilities
CP QoL–CHILD	Cerebral Palsy Quality of Life Questionnaire for Children
DISABKIDS CPM	DISABKIDS Cerebral Palsy Module
FCS	Family-Centered Services
FQL	Family Quality of Life
GBD	Global Burden of Disease
HICs	High Income Countries
HRQoL	Health Related Quality of Life
ICF	International Classification of Functioning, Disability and Health
JIA	Juvenile idiopathic arthritis
LMICs	Low- and Middle-Income Countries
PedsQL	Paediatric Quality of Life Inventory
QoL	Quality of Life
REC	Research Ethical Committee
ROM	Range of Motion
SCA	Standard Capability Assessment
VAS	Visual Analogue Scale
WHO	World Health Organization

Glossary

Term

Explanation

Applicability

The degree to which something, such as a measurement tool, can be applied in a particular context or population. It focuses on the practicality and feasibility of using a tool in a specific setting.

Appropriateness

The degree to which something is suitable, relevant, or fitting for a given context or population. It focuses on the alignment between a tool and the characteristics, needs, and values of the target population. It considers aspects such as cultural sensitivity, linguistic relevance, and the ability to capture the specific domains or aspects of quality of life that are important and meaningful within that population.

Community Based Rehabilitation

The World Health Organization explains it as: “a strategy within general community development for the rehabilitation, equalization of opportunities, and social inclusion of all people with disabilities”. It addresses the following five key aspects: health, education, livelihood, social and empowerment. It is realized through the collaboration of multiple parties, persons with disabilities, their families and communities, governmental and non-governmental organizations.

Community Health Worker

This paper has adopted the following statement: “A community health worker is a frontline public health worker who is a trusted member of and/or has an unusually close understanding of the community served. This trusting relationship enables the worker to serve as a link between health/social services and the community to facilitate access to services and improve the quality and cultural competence of service delivery.”

Family-Centred Service

CanChild’s definition: “Family-Centered service is made up of a set of values, attitudes, and approaches to services for children with special needs and their families. It recognizes that each family is unique; that the family is the constant in the child’s life; and that they are the experts on the child’s abilities and needs. The family works with service providers to make informed decisions about the services and supports the child and family receive. The strengths and needs of all family members are considered.”

Acknowledgements

Hereby I would like to express my sincere gratitude to all those who have contributed to the completion of this thesis.

First and foremost, I would like to express my gratitude to the thesis advisor, for the feedback and support I received. Throughout the entire process elaborative and extensive feedback was provided, which allowed me to grow and develop as a researcher and academic writer. The expertise of the thesis advisor played an immense role in the quality of the end result of my work.

Additionally, I wish to extend my appreciation to the academic advisor, whose guidance was of great importance during my academic journey. Persistent willingness to help and timely responses via email greatly facilitated the thesis process.

I want to offer my profound thanks to all the experts who participated in interviews. The willingness to share their valuable time, experiences, perspectives and insights greatly contributed to this research.

A special thanks to the entire KIT staff for high quality of knowledge acquisition during the master program. Inspiring and passionate lecturers nurtured my intellectual curiosity.

My appreciation goes to the entire NTC (Netherlands Course on Global Health and Tropical Medicine) class of 2020. For the rich discussions we had and the given support. Not to mention other inspiring students I came across during the advanced modules at other universities. I am indebted to ThankGod Zachariah Kums and Mare Romkes for their critical feedback and willingness to engage in numerous thought-provoking discussions.

Lastly, I am deeply thankful for the encouragement and love I received from my family and friends throughout this journey. Their belief in my abilities have been my driving force to stay focused and overcome challenges along the way.

1) Introduction

I graduated as a physiotherapist in 2018. My passion for International Health started during a bachelor's programme named 'healthcare professionals from an international perspective'. I had the honour of working closely together with the Red Cross and Home Based Care in Genadendal, South Africa. This experience was my initial exposure to working as a healthcare professional in a low resource setting, and it profoundly sparked my career aspirations. Seeing the challenges faced by individuals in underserved communities motivated me to contribute to the field of international health.

In March 2020, I started my academic journey at KIT in Amsterdam, which provided me with a valuable opportunity to further develop my knowledge and skills in the realm of international health. Combining physiotherapy and international health brings together principles and practices of physiotherapy with the global perspective and challenges of international health. This combination of health disciplines is important for improving healthcare outcomes and addressing the diverse needs of populations worldwide. Physiotherapists often engage in community outreach programs to promote health and well-being. By reaching out to underserved populations, physiotherapists contribute to improving public health and the reduction of health disparities.

Through a collaboration with KIT, the foundation Cerebral Palsy Africa emerged with a question that matched my previous experience and future goals. Cerebral Palsy Africa is a Dutch charitable incorporated organization working in four African countries to improve the lives of children with cerebral palsy. The organization strives to integrate these children into their communities and ensure their access to education. This is achieved through raising awareness about the benefits of therapy and conducting training programs for community workers, teachers, parents, and therapists. By equipping these individuals with knowledge and skills, cerebral palsy Africa aims to enhance the children's functional abilities in their everyday lives.

A collaboration was established between Cerebral Palsy Africa and myself. Cerebral Palsy Africa is in dire need for recommendations on what measurement tools to use to measure change in the quality of life, as a result of their care and rehabilitation interventions, in children with cerebral palsy. Such measurement tools provide necessary evidence on whether applied interventions are leading to improvements in the lives of the children with cerebral palsy and their family. Cerebral Palsy Africa is interested to find out what the best way is to measure the effects of the interventions that are applied by their partners. Therefore, this thesis will focus on quality of life measurement tools for children with cerebral palsy in low- and middle-income countries.

2) Background

Cerebral palsy (CP) is one of the major causes of childhood disability, it is a non-progressive lifelong condition.¹ It affects the activity and participation of children worldwide impacting their quality of life (QoL).¹ It is an umbrella diagnosis denoted as: “a group of permanent nonprogressive disorders of the development of movement and posture, causing activity limitation, that are attributed to disturbances that occurred in the developing brain”.² The motor disorders of CP are often accompanied by disturbances of sensation, perception, cognition, communication, and behaviour caused by epilepsy.² Secondary are musculoskeletal problems such as muscle contractures, bony torsion, hip displacement, and spinal deformity.² Two different groups have been classified, a pyramidal group including the motor cortex of the brain (e.g. spasticity) and an extra pyramidal group including the basal ganglia and cerebellum of the brain (e.g. dystonia and ataxia).³ To make a distinction within the pyramidal group subgroups are made, *hemiplegia* (unilateral upper and lower extremity) *diplegia* (bilateral lower extremity) and *quadriplegia* (all extremities involved).³

Research to identify the prevalence of CP has been mostly undertaken in high-income countries (HICs), through registers of child impairments or large population-based studies. Such registers are often not in place or incomplete in low- and middle-income countries (LMICs).⁴ However, a recent systematic review published new estimations showing that between January 2011 and November 2020, the prevalence of CP was approximately 0.2% (95% CI: 0.1–0.2) for HICs and 0.3% (95% CI: 0.3–0.4) for LMICs among children 0–18 years.⁵ It is estimated that globally, children with moderate to severe motor impairments have a global burden of disease (GBD) of 0.9% (95% UI: 0.8–2.0).⁵ In HICs, the GBD prevalence is approximately 0.6% (95% UI: 0.5–0.6) which indicates a higher prevalence for LMICs than the documented global estimate.⁵ Potential reasons include certain risk factors that are more present in LMICs such as low birth weight, premature birth, birth asphyxia, and infectious diseases during pregnancy.⁶ Furthermore, in LMICs early detection of a child with a disability and presenting it to healthcare services do not rely on routine screening by clinicians working in primary healthcare, but rather on the parents or caregivers. In these countries, considering the high burden of infectious diseases and poor accessibility of healthcare services, many disabilities of children are unrecognized or identified very late.^{6,7} Additionally, the improvements in the management of premature and complicated deliveries as well as better maternal and neonatal care are another contributing factor. This can be explained by the fact that all children in LMICs, (including children with cerebral palsy) have higher survival chances; i.e. the under -5 mortality rate decreased significantly in this group of countries.⁸ This causes neonates to reach an age where CP can be diagnosed.⁹ For the aforementioned reasons it is expected that the actual prevalence of CP is even higher than stated before.

Children with CP highly depend on rehabilitation services in order to maintain their health. Organizing healthcare management for children with CP can be complex, due to the multidisciplinary nature.⁶ In most of the HICs, allied health professionals such as physiotherapists, occupational therapists, nutritionists, and speech & language therapists are involved in the care of children with CP. Physiotherapy, in particular, plays a crucial role in the management of children with CP, focusing on function, movement and maximizing the opportunities for the child to reach its full potential.¹⁰ This can be challenging in LMICs where there are severe shortages of rehabilitation professionals and services.¹¹ Findings from

an international multi-center cerebral palsy register in Bangladesh, Indonesia, Nepal, and Ghana show that nearly half of the children notified in CP registers never received any form of rehabilitation services.¹¹ Because of the severe shortages of rehabilitation professionals in low-resource settings, children with CP mostly rely on family-centered services (FCS) and community-based rehabilitation (CBR). It is known that FCS is highly valued by parents and healthcare providers and seen as effective for the outcomes of the child and their family and service delivery.^{12 13} The primary focus of CBR programs is the improvement of QoL for people with disabilities and their families. Its goals are directed at the inclusion and participation of the child with CP in the family and community, it is context-based and individualized care.¹⁴ This approach can be difficult to implement because it involves training of family/community members to provide these services.¹⁴

Another challenge that presents itself in LMICs is financial hardship. Poverty intensifies challenges for children with CP in LMICs.¹⁵ For example, the high cost of medical services and geographical barriers hinder parents' access to services and make it difficult for them to seek help.¹⁵ This can result in unmanaged comorbidities including pain and epilepsy.¹⁵ In addition, inadequate feeding strategies combined with food insecurity, can result in malnutrition which leads to increased morbidity.¹⁵ Consequently, it is of great importance that children with CP receive interventions tailored to their specific needs.

Interventions for CP children can be evaluated in various ways. In order to measure well-being, QoL can serve as an outcome to evaluate the effect of interventions, whether applied by healthcare professionals or community health workers (CHWs).¹⁶ Measuring QoL has the potential to evaluate interventions more holistically because QoL is covering multiple life domains. According to Barcaccia et al. (2013), QoL has been used with too much inconsistency and has a wide variety of interpretations.¹⁷ There is an overall agreement that QoL is a multidimensional and a subjective concept.^{16 18} There seems to be a widely accepted agreement on the World Health Organization's (WHO) definition of QoL: "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."^{19,20} The QoL of children encompasses more than just their perception of social, physical, and emotional well-being, it should be adaptable to changes that occur as they develop.²¹ To ensure a clear and consistent outcome to measure QoL, it is essential to define QoL for the population being studied. The International Classification of Functioning Disability and Health (ICF) is widely used in healthcare by various disciplines and captures QoL domains.²² It is a framework that can be used to gather information on functioning and disability.²³ The ICF encompasses all aspects of potential factors that might have an influence on the QoL of children.²⁴ A study done by Fayed et al. (2012) analysed generic patient outcome reports on content and use of the WHO definition.²⁰ Out of 15 instruments being analysed, only four were recognized as having QoL perspectives using the WHO definition whilst 12 of them were frequently applied as QoL measures.²⁰ The fact that measurement instruments have been used wrongly in the past implies that there is a lack of understanding of QoL and/or there is a lack of instruments to measure QoL in children with CP.²⁵

3) Problem statement, justification and objectives

3.1 Problem statement

3.1.1 Measurement tools

It is of great importance to put emphasis on the original purpose of the QoL instrument Cerebral Palsy Africa (CPA) wishes to use, as it can significantly impact the direction and focus of the domains and items.²⁶ For example, the Paediatric Outcomes Data Collection Instrument (PODCI), Children Health Questionnaire (CHQ), and the Lifestyle Assessment Questionnaire (LAQ) have been used to measure QoL. However, these instruments are not developed with the goal to measure QoL.²⁶ The PODCI is designed to measure functional status, the CHQ is to measure functional status and well-being and the LAQ is to measure the impact of disability.²⁶ Using these measurement tools to evaluate QoL has substantial limitations.²⁵ A systematic review done by Makris et al. (2021) draws attention to the inconsistency of the conceptualization of QoL in paediatric CP, with a focus more on “ill-being” instead of “well-being”.²⁷

Existing measurement tools can be divided into two different groups:

- 1) Generic measures are developed to make an overall assessment of a person, it can be used in both sick and healthy people. This is more suitable for comparing QoL across different populations.^{28,29}
- 2) Disease-specific measures are specifically developed for patients with a particular disease or condition. It focuses on the concerns and issues relevant to that specific population.²⁹ For example, patients’ symptoms, difficulties, or comorbidities of a certain disease.³⁰

Disease specific measurement tools have not received the same attention and development as generic measurement tools and are often not free of charge.³⁰ Disease specific instruments measuring QoL in children, specifically children with disabilities, lag even more behind. This is primarily because of the particular measurement challenges associated with paediatrics e.g., autonomy of children, continuous developmental changes, choosing which domains to measure, and deciding whose perspective to include.²⁸ Most of the disease-specific instruments developed for measuring QoL in children with CP have been developed in HICs.^{26,31} For example, the research team that developed the Cerebral Palsy Quality of Life Questionnaire for Children (CP-QoL CHILD) consisted of experts solely from HICs namely Australia, the United States, Germany, and Scotland.²⁹ In addition to that, the qualitative interviews (that provide input into the instrument) were held with participants from the Victorian Cerebral Palsy Register maintained by the Department of Child Development and Rehabilitation at the Royal Children’s Hospital Melbourne in Australia.²⁹

Additionally, disease specific measurement tools can capture important aspects of daily life that are unique to this patient group.²¹ Disease specific CP measurement tools or generic measurement tools with a disease specific CP module are able to explore a child’s feelings for example about therapeutic interventions, adaptive equipment and social participation.³² In contrast to generic measures, disease specific instruments are also more capable to measure changes that are relevant to health professionals and patients.^{16,21,30} This paper will investigate measurement tools that evaluate the effect of interventions and measure change in QoL. For this reason, this paper will refrain from including questionnaires that simplify QoL into a single score. Instead, it will exclusively focus on disease specific measurement tools or generic measurement tools with a disease specific module.

3.1.2 Culture and context

Despite recent progress, challenges persist in achieving international comparability and incorporating cultural aspects in the development of QoL instruments for children.³³ It would be incorrect to believe that QoL dimensions (used in questionnaires) developed in HICs are similar and directly comparable to LMICs.³⁴ To utilize a questionnaire developed in another setting, a technique called cross-cultural validation can be employed.³⁵ Kagawa-Singer et al. (2010) express their concerns about the cross-cultural validation technique involving translation, back translation, and reviewing.³⁴ If the initial concept of the instrument or the original version is not tested for cross-cultural validation, it might not be satisfactory enough.³⁴ Literature that identifies the concept of QoL for children with CP in LMICs is scarce.³⁴ Qualitative studies support the idea that QoL varies among cultural groups. For instance, diverse cultures have different beliefs about the concepts of “health” and “sickness”, and interpretations of bodily symptoms are highly influenced by culture.³⁴ East African cultural beliefs about the causes of disability are divided into three distinct groups by S.G. Harknett.³⁶ Firstly, traditional animism includes beliefs that disabilities are punishments for bad actions that happened in the past, including witchcraft exercised by other people.³⁶ Secondly, Christian fatalism includes beliefs that having a child with a disability is the will of God.³⁶ Lastly, medical determinists believe that disabilities are a consequence of modern medicine.³⁶

A related reason why the use of measurement instruments developed in HICs can be questioned for use in LMICs is that there is also a lack of recognition of the influence of American and Northern European cultures on the concept of QoL.³⁴ This has impeded valid cross-cultural comparisons of QoL globally. This can be attributed to the notion of QoL and its underlying values being universally applicable, as well as the lack of qualitative studies to capture the perceptions and definitions of alternative cultural groups.³⁴

3.2 Justification

As there is no cure known for cerebral palsy, QoL is an important outcome to assess the effect of interventions.²⁷ A study researching the effectiveness of child health interventions in LMICs shows that previously, only intermediate clinical markers were used.³⁷ Examples are measurement of Range of Motion (ROM) of a joint, muscle strength and pain scales such as the Visual Analog Scale (VAS). Thus QoL as a holistic notion of well-being has the potential to measure effects from a broader perspective.³⁸ This holistic perspective is particularly important for children with CP because the severity of CP does not automatically correlate with QoL.²⁷ This is also known as the ‘disability paradox’.³⁹ A qualitative study by Albrecht and Devlieger 1999 reports that 54.3% of persons with moderate to serious disabilities stated that they had an excellent or good QoL.³⁹ These numbers confirm that the disability paradox exists.³⁹

After the United Cerebral Palsy Association (1991) included the objective of enhancing the QoL of individuals affected by CP in their mission statement, QoL received more attention. This has resulted in QoL becoming the foremost outcome of interventions for children with CP.⁴⁰ Information on QoL is valuable for policy planners at every level, and for health professionals it is crucial to demonstrate the impact of their interventions.¹⁶ For this reason CPA, is interested in using a measurement tool that evaluates QoL in children with CP in LMICs. In order to measure the impact of the interventions applied, CPA must face a critical imperative in determining which measurement tool(s) to employ. They want to make sure that their initiatives effectively bring about essential improvement in the lives of children with CP

and their families. The aim of this study is to investigate the applicability and appropriateness of existing measurement tools that evaluate QoL in children with CP in LMICs, to provide recommendations on which measurement tools to use.

3.3 Objectives

The overall objective of this study is to assess the applicability and appropriateness of existing measurement tools to evaluate QoL in children with CP in LMICs. The following sub-objectives have been formulated:

- To identify the definition of QoL for children with CP living in LMICs.
- To assess which domains of QoL should be considered in children with CP living in LMICs.
- To identify strengths and weaknesses of existing QoL tools that evaluate QoL in children with CP living in LMICs.
- To gain a comprehensive insight into experts' perceptions of the use of existing QoL measurement tools on children with CP living in LMIC.
- To provide recommendations for CPA about the type of measurement tool that best can be used in evaluating the effect of interventions in children with CP living in LMICs.

4) Methodology

4.1 Study design

In order to understand experts' experiences a qualitative research design was chosen. In this design the phenomena are explored by including the perceptions of research participants to address the research question.⁴¹ The research methodology is based on a phenomenological approach with inductive reasoning.⁴² The purpose of having phenomenology as a theoretical framework is to gain an in-depth understanding of a person's subjective experience and perspective of a certain phenomenon.⁴² Phenomenology allows the researcher to gather expert perspectives on the applicability and appropriateness of existing measurement tools used to measure QoL in children with CP in LMICs. An extensive literature review was performed in order to gather data on the definition and domains of QoL for children with CP in LMICs. Furthermore, the literature search was employed to identify strengths and weaknesses of existing QoL measurement tools. Thereafter semi-structured in-depth interviews were conducted by one researcher (BS) with the guidance of a topic guide. After transcribing the interviews, reflexive qualitative thematic analysis was performed in order to analyse the data. Categories emerged from this, and categories were grouped into themes and written down in the findings.

4.2 Participants

For the semi-structured in-depth interviews, participants were selected through purposive sampling – expert sampling. A set of criteria was developed and used to select eligible participants. Healthcare professionals with at least five years of working experience with CP children in LMICs were selected. The thesis advisor was used as a contact person for the researcher and provided the researcher with a list of potential experts. For maximum variation, experts from different allied health disciplines were sampled. With the aim of reaching higher degrees of saturation, this study intended to include 11-12 participants.⁴³ Literature indicates that the response rate for participating in interviews is approximately 30%.⁴⁴ Therefore, 32 experts were invited to participate in the semi-structured in-depth interviews. This included 14 physiotherapists, seven occupational therapists, two speech and language therapists, three paediatricians, three researchers, and one midwife. They were approached via email; the invitation can be found in Appendix 1. This email included a brief introduction of the content of the study, its study objectives and an informed consent form which had to be signed and returned by the participants. The informed consent form can be found in Appendix 2. After the invitation was sent, 10 healthcare professionals accepted the invitation and gave informed consent to participate in the study. One participant was selected on the basis of a recommendation made by an invited healthcare professional.

4.3 Data collection

4.3.1 Literature review

The review was based on a literature search through Google Scholar. The keywords 'quality of life' OR 'health related quality of life' AND 'children with cerebral palsy' OR 'children with neurodevelopmental disability' OR 'neurodevelopmental disorders' AND 'disease specific measurement tools' were used. Studies were included if children were between the age of 0 and 18 yrs. Articles published between January 2001 and 31 December 2022 written in English language and full text available were selected for consideration. Thereafter, titles and abstracts were screened for eligibility guided by the inclusion criteria listed before. The snowballing technique was applied to include relevant articles that were potentially missed during the initial search.

4.3.2 Semi-structured in-depth interviews

A topic guide with 21 questions was developed to navigate the semi-structured in-depth interviews. The topic guide can be found in Appendix 3. To test the topic guide, a pilot trial was held, two pilot interviews were organized to make sure that the questions were clear and relevant to the research question. It was an iterative process, and in response to the given feedback questions were revised. The data analysis did not incorporate the two pilot interviews. Before every interview, the interviewee received a short introduction letter with a summary of the study and its objectives. The day before the interview, a reminder was sent to the participant, which included the interviews topic guide. This was done to allow the participant to become familiar with the topic and to prepare for the interview if wished. The interviews had a conversational basis and the duration of the interview was approximately 40 minutes. All interviews were held in English remotely through Zoom; a cloud-based video conferencing service. The researcher conducted all interviews alone. After consent was given by the participant, an audio recording was done with the goal of transcription afterwards. Artificial Intelligence (AI) provided by Zoom was utilized to support the transcription process of the recorded interviews. While AI served as a valuable time-saving tool, human validation and review were undertaken by the researcher to ensure data integrity and to correct potential errors. Following the transcription process, the outcome was sent back to the participants giving them the opportunity to correct and provide additional feedback. Finally, after agreement of the participants the transcripts were analysed. Interviews were planned and conducted until the end of June 2023.

4.4 Data analysis

4.4.1 Analysis of literature

The included studies were evaluated based on content. The researcher screened the studies by title and abstract for eligibility. Articles with ongoing uncertainty regarding eligibility received full-text were reviewed by the researcher. Identified studies were retrieved in full-text and data extracted including study aim, location of study, strengths and weaknesses of the measurement tool, psychometric properties, cross-cultural validation, participant inclusion and exclusion criteria, outcomes, and results.

4.4.2 Analysis of semi-structured in-depth interviews

Semi-structured in-depth interviews were transcribed by intelligent transcription to capture the essence of the spoken language. After transcription, reflexive qualitative thematic analysis was performed in order to analyse data; this was done by Braun and Clarke's approach.⁴⁵ The coding process involved two stages. First, the researcher conducted initial coding to become familiar with the data set. Subsequently, a line-by-line coding approach was used to refine and expand upon the initial codes. Because participants came from different cultural backgrounds it can be difficult to accurately infer meaning. Therefore, inductive in vivo coding was applied, with the goal to stay as close as possible to the original words and phrases without interpretation from the researcher. Atlas.ti software was used to code the transcribed data. The code tree can be found in Appendix 4. Categories emerged from this, and categories were grouped into themes and written down in the findings. After data analysis, the qualitative data was informed by consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups.⁴⁶ Subsequently, the results section integrated findings from both the literature search and the semi-structured in-depth interviews.

4.4.3 Rigour

A foreseen limitation was sampling bias. In order to avoid personal biases, clear selection criteria were established based on the research question and objectives. This helped to ensure that the selection of participants was based on relevant characteristics and not on personal preferences. Participants' feedback was invited to confirm that the experts' real perceptions were captured. However, only two participants provided feedback.

4.5 Ethics and integrity

An ethical waiver was obtained through the research ethical committee (REC) of the KIT Royal Tropical Institute. In Appendix 5 the ethical waiver can be found.

5) Results

This section presents the findings of this study integrating both the results of the literature review and the semi-structured in-depth interviews. Through qualitative analysis four overarching themes were identified which will serve as the guide for this section. The themes are: 1) Conceptualizing QoL, 2) Key QoL domains, 3) Existing QoL measurement tools, and 4) Experts' insights on tool usage for CP children in LMICs. For each theme, an overview of relevant literature is provided followed by experts' perspectives in accordance with the aforementioned themes.

5.1 Characteristics experts

In the study a total of 11 experts participated including, 6 physiotherapists from whom 4 were specialized in children's physiotherapy, 1 paediatrician, 1 medical specialist in rehabilitation medicine, 1 clinician, 1 speech therapist, and 1 occupational therapist. To maintain anonymity each expert was assigned to a unique expert identity number. Table 1 shows an overview of expert details.

Expert	Profession	Years of experience	Countries
X1	Physiotherapist	17 years	Zambia and Malawi
X2	Developmental paediatrician	50 years	Colombia, Mexico, Chile, India, Pakistan, Sri Lanka, Israel, Iceland, Kenya and South Africa
X3	Physiotherapist	30 years	Malawi, Bolivia and Zimbabwe
X4	Physiotherapist, specialized in children	10 years	Ghana
X5	Speech therapist & nutrition specialist	40 years	Cameroon, Ethiopia, Vietnam, Ghana, Philippines and Vietnam
X6	Physiotherapist, specialized in child development	10 years	Rwanda
X7	Medical specialist in a rehabilitation medicine	30 + years	Vietnam, Tanzania, El Salvador, Chile, Sri Lanka, Indonesia, Laos, Afghanistan, Costa Rica, Rwanda, Nepal, Bangladesh, Philippines and India.
X8	Physiotherapist, specialized in children	25 years	China, South Africa, Romania, Bulgaria, Malawi, Mozambique, Kenya, Ghana, Sri Lanka, India, and North Korea
X9	Clinician	6 years	Uganda
X10	Occupational therapist	22 years	South Africa and Madagascar

X11	Physiotherapist, specialized in children	18 years	Zimbabwe, Ghana and Jordan
------------	--	----------	----------------------------

Table 1 overview experts details.

5.2 Conceptualizing QoL

Literature

Waters et al. (2004) conducted qualitative research to find out what QoL means for children with CP. Interviews held with children diagnosed with CP and their parents in relation to their QoL yielded valuable insights into their daily experiences.^{25,31} Specific themes unique to children with CP emerged such as bodily pain, daily tasks, communication, and future QoL.^{25,31} This study included children from the Victorian Cerebral Palsy Register (from HIC), the meaning of QoL for children with CP in LMICs is not covered in the literature.

Experts perspectives

According to most of the experts, the definition of QoL was described as subjective to each child and the inclusion of the family is indispensable. It differs from children residing in HICs because culture and context influence people's perspectives on QoL. It was explained as a complex and value-laden definition, including different domains which will be expanded on in the second theme. Experts reflected on the definition of QoL for children with CP in LMICs as follows:

X1: "So for me, quality life is not a one size fits all sort of thing. It has to be tailor-made for every child in every family."

X2: "I don't know what quality of life means. I don't know what it means to you, let alone to children with cerebral palsy in low- and middle- income countries. I don't often know what it means in our own community, because people have so many different meanings for the term quality of life."

X6: "But I think the quality of life it's a very complex concept to measure. Now, as we said, it can be subjective so we need to be a little more contextualized.."

There was an overall agreement that QoL is an encompassing important key outcome that can be used to measure the impact of interventions. Clinical single markers often fail to demonstrate improvements in children who are diagnosed with severe CP. These improvements are not solely limited to a greater ROM of a specific joint or reduction of spasms. Instead, it can manifest in higher levels of participation for the child. To exemplify, a child supported by walking aid gains more independence, allowing the mother to attend to other household chores while the child is standing safely in the walking aid. This example can improve the QoL for both the child and the family. Because of its comprehensive nature, QoL captures multiple domains of life. Experts highlighting the importance:

X4: "I think the concept of quality of life is all encompassing, and I think it's still because it has different domains. I think it's a good way to assist the well-being of the child. So maybe for a lower middle income country. It's really looking at how to make it more culturally and contextually relevant, how to capture the other things that are unique in our environment. And to make it make more sense for us, culturally. Yeah. But I still think it's an important way to go."

X9: “Actually, I agree, it's a good measure to look at, because I mean, this is the thing we can fix for our children that we are looking after. And I think if we don't.. the best way to come out strong is measuring all the different aspects in their lives, so that we can enforce the systems in order to set up .. adjust the factors which impact on that well-being of the people with cerebral palsy. And this can only be measured by using the quality of life.”

5.3 Key QoL domains

Literature

The literature search yielded no results regarding QoL domains for CP children living in LMICs.

The perspectives from experts

Experts identified the following QoL domains: 1) Beliefs & Stigma, 2) Family, 3) Resources & Access 4), Education and 5) Food & Malnutrition. The subsequent section will expand further on the emerged themes. A detailed description of the theme will be provided, accompanied by quotes from the interviews held with the expert panel. Figure 1 includes quotations made by the experts to illustrate their opinions on the emerged domains.

5.3.1 Beliefs & Stigma

The first and largest domain is ‘Beliefs & Stigma’, this was mentioned 36 times. Beliefs about children with disabilities have a great impact on their QoL. Being accepted and having quality relationships with people in the community play a vital role in African cultures. In some African communities, there is a belief that having a child with a disability is a punishment or a curse to the family, which stems from ancestors many years ago. It is seen as the result of a sinful act committed by one of the parents. Some cultures hold the belief that it has something to do with witchcraft, which causes delays in presenting the child to healthcare services. Because parents feel angry, ashamed, and scared at times this can also lead to harmful practices to “fix” the child. According to the interviewed experts, children with CP rely heavily on support from the family and community. Hence, it is important that they can grow up in a welcoming environment free from negative beliefs and misconceptions about the origin of cerebral palsy.

5.3.2 Family

The second category that emerged was ‘Family’, mentioned 30 times by the experts. It appeared that the involvement of parents and caregivers is essential to children with CP. There seems to be an inevitable link between the well-being of the family and the well-being of the child. Family is fundamental in most cultures. Inclusion and support from family influence the QoL of children with CP. The family can facilitate inclusive practices for the child within the family but also in the broader community. Expert X10 highlighted the importance of including the family with an example of her experience.

X10: “I mean often our kids with CP are born to very young mothers, and sometimes, for example, they are newly married in a household and in a lot of our communities. When a woman gets married she spends the first 2 or 3 years in rural areas with quite a traditional culture. She'll spend the first few years living with her mother-in-law and she must work extremely hard, physically extremely hard to prove herself to her new family. So she's carrying the water, and she's carrying the wood, and she's working in the garden, and she's, you know, she's really busting a gut to prove herself. And telling that mom that she must take time to do exercises with her child, or feed her like this, or do like that. It's not gonna work.

So you can, you know, you can educate them until you are blue in the face about what's good for the child. But unless you're going to sit down with the whole family, and particularly the mother-in-law, and engage in all of that, and deal with the whole family situation. (...) But basically quality of life should look at the household, not just the child, because you can't separate them out."

5.3.3 Resources & Access

The third main category mentioned was 'Resources & Access', mentioned 21 times. The interviewed experts asserted that children with CP in LMICs have less access to resources as compared to children in HICs. They state that the QoL of children with CP is influenced by the access, or the lack of it, which they have to resources such as healthcare, shelter, food, and housing. To illustrate this; an adapted spoon or a walking aid can make a significant difference in the lives of children with CP, enhancing their independence and mobility. Another given example is the level of access to physiotherapy. In some countries where experts have worked, physiotherapy is considered a luxury. Several experts stated that there are children in LMICs who die at a young age and never saw a physiotherapist in their lives.

Another point highlighted by expert X3 was that in some rehabilitation centers, the rehabilitation assistants use outdated techniques. Therapy is mainly focused on hands-on techniques such as massage or passive range of motion exercises. With hands-on techniques, they try to improve their muscle flexibility and joint range of motion. Mostly this is very painful for the children and the desired result is not forthcoming. The health workforce at rehabilitation centers including rehabilitation assistants lack access to the latest available knowledge. The usual lack of educational resources results in inefficient working techniques and ultimately impacts the highest achievable health outcomes for children with CP. Even though CP is a non-progressive condition, it has progressive development of various clinical manifestations, including musculoskeletal consequences. Therefore physiotherapy continues to be of utmost importance throughout children's lives. It plays a vital role in managing CP-related comorbidities and preserving their overall well-being.

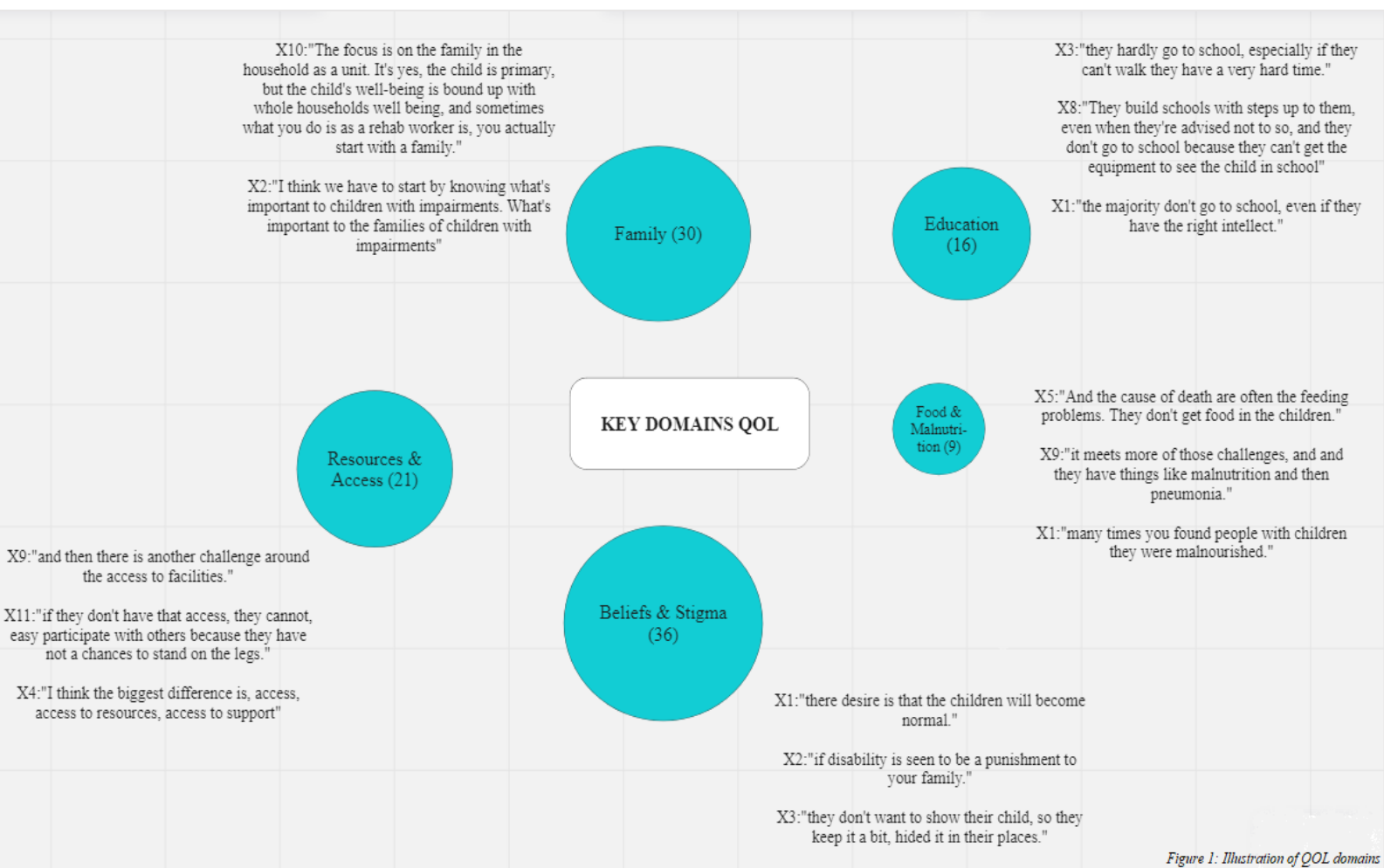


Figure 1: Illustration of QOL domains

5.3.4 Education

Fourth, the experts addressed 'Education', for improving the QoL of children with CP, mentioned 16 times in total. As reported by experts, half of the children with CP experience learning difficulties, while the other half do not encounter such challenges. This corresponds with conclusions from literature that indicate that half of the children with CP have an average or higher-than-average IQ.⁴⁷ Despite the right intellectual capacity to attend school, schooling – for various reasons - is often not facilitated for them. As an example, there are teachers unwilling to take children with disabilities into their classes. Other children may engage in bullying children with disabilities. Another barrier mentioned by one of the experts is the challenge the children have in going to the toilet independently. If the child is unable to do so, they are usually not permitted to attend school. These challenges pose barriers for children with CP to access education. As a consequence, children with CP do not have equal opportunities as their peers, limiting their ability to develop themselves. In HICs, there are many stories of adults diagnosed with CP who have gone up to PhD level, whereas children in LMICs often drop out of school before secondary school.

5.3.5 Food & Malnutrition

The fifth and last category is 'Food & Malnutrition', mentioned nine times. Experts shared that mothers or caregivers encounter challenges with feeding their children with CP. They are not able to effectively nourish the child, potentially leading to malnourishment. Consequently, the child lacks sufficient energy for proper development. Another consequence of malnutrition mentioned by experts X5 and X9 is pneumonia. Expert X5 emphasized that parents and caregivers encounter the difficulty of physically choking when feeding their child. This is mostly because the child is positioned in the wrong way. The majority of the experts shared that children with CP in LMICs often spend time laying on the ground.

Community healthcare workers lack knowledge on how to address such challenges. One of the nutritionists shared that feeding strategies for children with CP have received more attention thanks to the efforts of UNICEF and WHO.

To demonstrate the data distribution across different disciplines and domains, a data stratification was performed. The findings revealed that the domain "Beliefs & Stigma" was mentioned by 10 experts, while both "Resources & Access" and "Family" were cited by 9 experts each. "Education" was mentioned by 5 experts, and "Food & Malnourishment" by 4 experts. Figure 2 visually presents the data stratification, showcasing the distribution of domains as cited by the different experts.

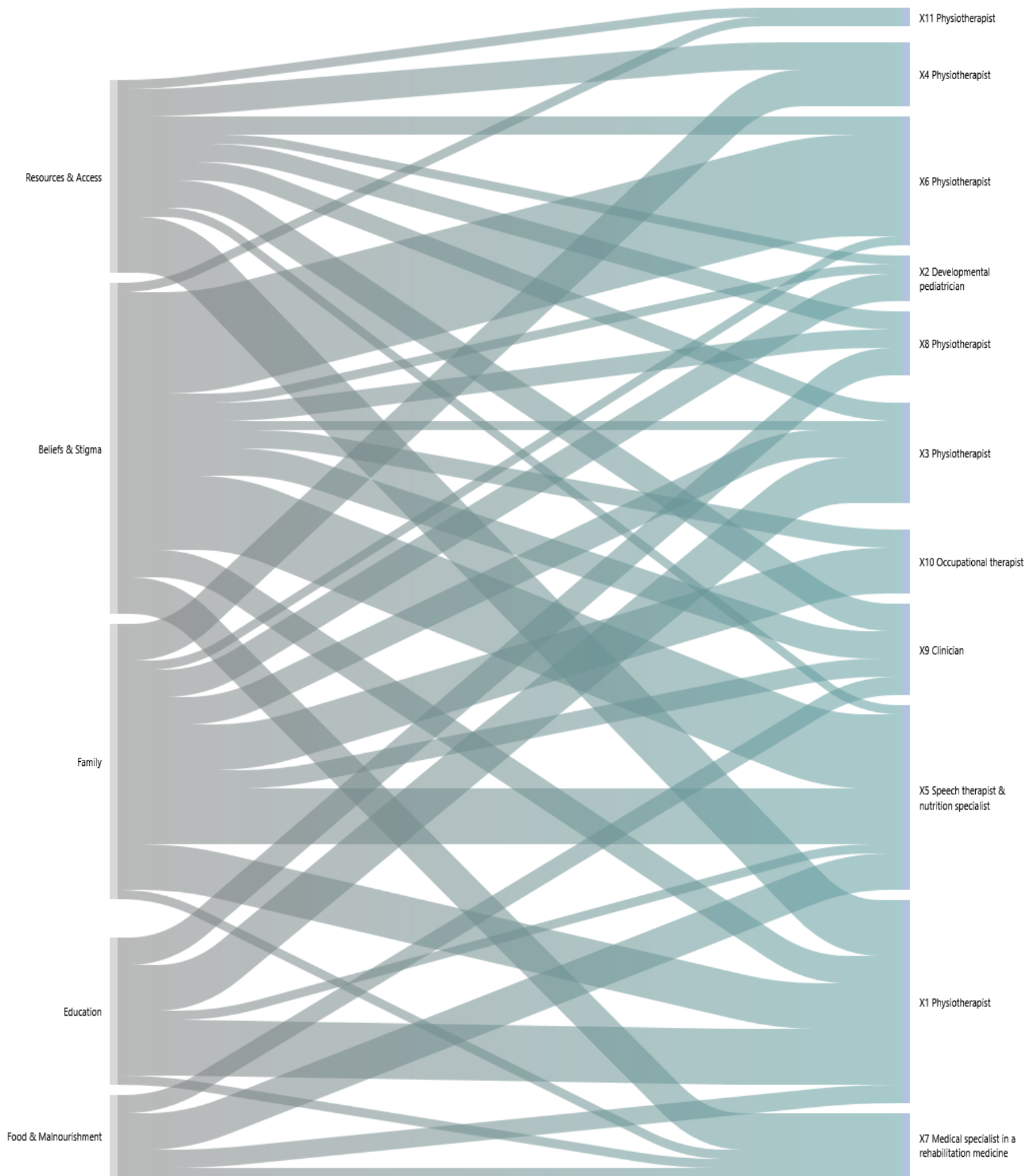


Figure 2 data stratification, distribution per QOL domain.

5.4 Existing tools

In this section, strengths and weaknesses of existing QoL measurement tools are described based on relevant literature followed by experts' opinions. Tools were selected based on the following inclusion criteria: disease specific measurement tools, or generic measurement tools with a disease specific CP module and originally developed to measure QoL.

Four disease specific measurement tools have been identified, CP-QoL CHILD, Peds-QL 3.0 CP, CP-CHILD, and the DISABKIDS CPM.

The CP QoL-CHILD was developed in 2004 by a team of researchers in Australia. It was the first questionnaire designed for children with CP that measures well-being.⁴⁸ The CP QoL-CHILD tool is seen as one of the strongest measurements because it is the only measurement tool that covers seven domains based on the ICF.³² Two versions are available a child self-report and a proxy parent report.⁴⁹ The child self-report consists of seven domains incorporated in the following chapters family and friends, participation, communication, health, special equipment, pain and bother, final questions.⁴⁹ The proxy parent report is identical to the child self-report but consists of two extra chapters: access to services and your health.⁴⁹

The PedsQL 3.0 CP module was developed for measuring health related quality of life (HRQOL) in children with CP.⁴⁷ It consists of formats for child self-report and parent proxy-report. Child self-report covers ages 5-18 years, while parent proxy-report covers ages 2-18 years it evaluates parental perceptions of the child's HRQOL.⁴⁷ The PedsQL 3.0 CP was developed by focus groups discussions, cognitive interviews, pretesting, and field testing protocols.⁵⁰

The CP-CHILD is a disease specific measurement instrument designed to measure caregivers' perception of health related quality of life in children with severe CP.²⁹ CP-CHILD is validated for caregivers of children with severe CP categorized in level IV or V of the Gross Motor Function Classification System.^{29, 1} The CPCHILD is developed based on recommendations of parents and caregivers, experienced health care professionals managing children with severe CP and by reviewing other questionnaires.²⁹

The DISABKIDS group has developed a European health related quality of life measurement tool for children and adolescents with a chronic medical condition and their parents.⁵¹ The project is a collaboration of the following seven countries (Austria, France, Germany, Greece, the Netherlands, Sweden and the United Kingdom) and included seven chronic medical conditions among them CP.⁵¹ The DISABKIDS CP Module consists of 10 items on two scales (impact and communication).⁵² The items on the two scales refer to difficulties related to functioning and activities of daily living both physically and socially.⁵² An overview of the four measurement tools and their main features is presented in Table 2.

Tool	Summary	Versions	Domains
CP-QoL CHILD	<ul style="list-style-type: none"> The CP QoL-CHILD was designed to assess the QoL of children with cerebral palsy aged 4-12 years.²⁹ Items are based on the ICF framework.²⁰ 	<ul style="list-style-type: none"> CP-QoL CHILD Primary Caregiver 4 – 12 years.²⁹ CP-QoL CHILD Self Report 9 – 12 years.²⁹ 	In total 66 items on 7 domains: <ul style="list-style-type: none"> Social wellbeing & acceptance Feelings about functioning Participation & physical health

¹ Gross Motor Function Classification System is a standardized system used to classify and describe the gross motor function of children and adolescents with cerebral palsy.³⁸

			<ul style="list-style-type: none"> • Emotional wellbeing & self-esteem • Access to services • Pain & impact of disability • Family health ²⁹
Peds-QL 3.0 CP	<ul style="list-style-type: none"> • The PedsQL 3.0 CP Module was designed to measure HRQOL dimensions specific to CP. • The questions and items of the PedsQL 3.0 was developed by focus groups, research and clinical experience.³² 	<ul style="list-style-type: none"> • Child self-report includes ages 5 to 7 years, 8 to 12 years, and 13 to 18 years.⁵⁰ • Parent proxy report includes ages 2 to 4 years, 5 to 7 years, 8 to 12 years and 13 to 18 years.⁵⁰ 	<p>In total 36 items on 7 domains:</p> <ul style="list-style-type: none"> • Daily Activities (9 items) • School Activities (4 items) • Movement and Balance (5 items) • Pain and Hurt (4 items) • Fatigue (4 items) • Eating Activities (5 items) • Speech and Communication (4 items)⁵⁰
CP-CHILD	<ul style="list-style-type: none"> • An instrument for children with severe CP examining health status, comfort, wellbeing and ease of caregiving.²⁹ • It was developed based on recommendations from caregivers, healthcare providers, and by reviewing of other measures (CHQ).⁵³ 	<ul style="list-style-type: none"> • CP CHILD proxy report. 	<p>In total 36 items on 6 domains:</p> <ul style="list-style-type: none"> • Personal Care (eight items) • Positioning, Transfer, and Mobility (eight items) • Communication and Social Interaction (seven items) • Comfort, Emotions, and Behaviour (nine items) • Health (three items); and • Overall Quality Of Life (one item).⁵³
DISABKIDS CPM	<ul style="list-style-type: none"> • The DISABKIDS family of questionnaires includes a set of developmentally appropriate and cross-culturally comparable questionnaires for assessing health-related quality of life in children and adolescents between 8 and 18 years of age.⁵⁴ • The items focus on the perceived impact of the child's well-being and not 	<ul style="list-style-type: none"> • DISABKIDS CPM 	<p>In total 10 items on 2 domains:</p> <ul style="list-style-type: none"> • Impact • Communication <p>Both scales include challenges concerning functioning, activities of daily living (physical and social functioning).⁵⁴</p>

	on performance of certain tasks, or functioning. ⁵⁴		
--	--	--	--

Table 2 overview general features

Strengths and weaknesses according to literature

The CP-QoL CHILD has three outstanding strong points. First, in the development of the tool, an international multidisciplinary collaboration took place. Second, items are based on the ICF framework. Third and last, the CP-QoL CHILD is the only measurement tool that focuses on well-being instead of ill-being. Literature also reveals challenges associated with the CP-QoL CHILD. A qualitative study conducted by Parkinson et al. (2010) highlights that children are not always familiar with the language used in the tool.⁵⁵ For example, the CP-QoL CHILD uses the wording: “being accepted”. The children that were interviewed in this study used words like “fairness” and not being “left out” and they mentioned being “picked on” or “bullied” by their peers.⁵⁵ In addition to that, also words such as “academically”, “communicate”, “independently”, “recreational activities”, “social events”, and “participate in your community” were used in the CP-QoL CHILD and not by the children participating in the study.⁵⁵

The Peds-QL 3.0 CP is based on qualitative research derived from focus group discussions and interviews.³² However, the included domains do not seem to encompass all important domains relating to the QoL of children with CP.³⁷ Adding to that the PedsQL 3.0 CP has a strong focus on functional outcomes for a measurement tool that measures QoL. Of the 35 items in the 13-18 year old parent report 23 items are about functional tasks.³²

The development of the CP-CHILD was based on the recommendations from caregivers, health care professionals experienced in the management of children with severe cerebral palsy, plus reviewing other questionnaires.⁵³ A limitation of the questionnaire is that the outcomes may not always reflect the child’s experience accurately, because it relies solely on caregiver reports.⁵³ Caregivers might misinterpret their child’s level of pain or discomfort, potentially leading to discrepancies in the reported data.⁵³

For the evaluation of interventions, the DISABKIDS CPM seems to hold great potential, because of its good discriminative ability.⁵⁴ However, the DISABKIDS CPM also has its shortcomings. Questions are phrased using negative language to assess the impact of disability.²¹ The questions are steered in such a way that it can threaten the self-esteem of young children by making assumptions.²¹ Examples are: ‘Is it frustrating to be unable to keep up with other children?’ ‘Do people think that you are not as clever as you are?’.²¹ Despite the fact that these items might have sound psychometric properties, there are possible ethical implications for the inclusion of such questions.²¹ In Table 3 a more detailed overview can be found of strengths and weaknesses of existing QoL measurement tools according to the literature.

QoL measurement tool	Strengths	Weaknesses
CP-QoL CHILD	<ul style="list-style-type: none"> Developed by an international multidisciplinary team of clinical and child health researchers in collaboration with parents and children with CP.²⁹ 	<ul style="list-style-type: none"> The words that being used in the CP-QOL CHILD are not specifically tailored to children.⁵⁵ Five children were included during the development; this might not have been

	<ul style="list-style-type: none"> • Items are based on the ICF framework.⁵⁶ • Measures well-being instead of ill-being cover six domains of QoL.^{56,48} • Time to administer 15-25 minutes.⁴⁸ 	<p>sufficient to reflect the voices of all children aged 9-12 years.⁵⁵</p> <ul style="list-style-type: none"> • Does not include items about the child's safety.⁵⁵ • The sensitivity is not yet established.²⁶ • No items about: "parents and extended family members, restful recreational activities and possessions, relaxing, tiredness, negative emotions or safety".⁵⁵ • Origin of the country: UK⁵⁶
Peds-QL 3.0 CP	<ul style="list-style-type: none"> • The questions and items of the PedsQL 3.0 were developed based research and clinical experience.³² • It is translated and culturally adapted in 70 different languages and cultures.⁵⁷ • Time to administer around 5 minutes.⁵⁰ 	<ul style="list-style-type: none"> • Qualitative data were used and derived by focus groups and interviewing, unfortunately details of the group participants were not provided.³² • It does not contain QoL domains that are included in most other tools for example: health perception, coping and adaptation, pain and discomfort.³⁷ • Non ambulatory children with severe cerebral palsy may find that several items in the Peds-QL 3.0 CP are not relevant to their situation. Aspects important to them impacting their QoL are not included in the questionnaire.²⁹ • Peds-QL 3.0 CP heavily focus on functional outcomes.³²
CP-CHILD	<ul style="list-style-type: none"> • It is a reliable and valid proxy measure of caregivers' perspectives on the health status, functional limitations, and well-being of children with CP.⁵³ • Developed based on the recommendations from caregivers, health care professionals experienced in the management of children with severe cerebral palsy, plus reviewing other questionnaires.⁵³ 	<ul style="list-style-type: none"> • Outcomes of the CPCHILD may not always reflect the child's experience correctly because it relies solely on caregiver reports.⁵³ Caregivers might misinterpret their child's level of pain or discomfort.⁵³ • The CPCHILD was initially developed in English language and thereafter validated in North American populations.⁵³ • As an evaluative tool for therapeutic interventions the responsiveness has to be established in future research.⁵³ • Time to administer 20-30 minutes.²⁹
DISABKIDS CPM	<ul style="list-style-type: none"> • It can be used in two different ways, for treatment evaluation and group comparison in clinical studies of children and adolescents with CP.⁵⁴ • It also has a good discriminative ability.⁵⁴ • The DISABKIDS family of questionnaires includes a set of developmentally appropriate and cross-culturally comparable questionnaires for assessing health- 	<ul style="list-style-type: none"> • Items are sometimes steered in a negative way which may threaten the self-esteem of children.²¹ • Age limit, starting from 8 yrs. • Included patients were sampled only from HICs: Austria, Germany, Greece, and United Kingdom plus their main caretaker.⁵⁴

	related quality of life in children and adolescents between 8 and 18 years of age. ⁵⁴	
--	--	--

Table 3 overview strengths and weaknesses existing measurement tools.

A study carried out by Waters et al. (2009) has pinpointed 7 crucial parameters essential for QoL measurement tools.²¹ In total 4 QoL measurement tools have been compared according to these parameters namely, CP-QoL CHILD, PedsQL, DISABKIDS-CPM, and CPCHILD. The CP-QoL CHILD emerges as the strongest measurement tool demonstrating excellent scores in 5 out of 7 parameters. It is notable that among the four measurement tools, three were originally designed to measure QoL, whereas the CP-QoL CHILD exclusively assesses well-being. Another noteworthy aspect is that all measurement tools score average in terms of their psychometric properties. Figure 3 presents an overview made by Waters and colleagues on how the 4 measurement tools are scored according to the 7 parameters.²¹

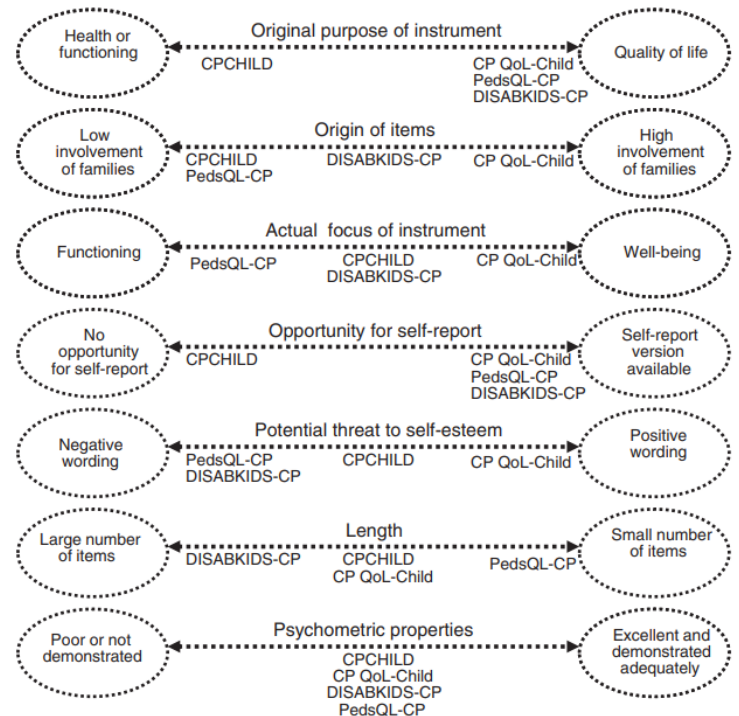


Figure 3 scoring CP CHILD, CP-QoL CHILD, PedsQL, DISABKID.

Strengths and weaknesses according to experts

There were 24 codes generated in the category: 'Strengths existing tools', against 51 codes in the category: 'Weaknesses existing tools'. The majority of healthcare professionals did not use QoL measurement tools in clinical settings. The main reason mentioned was because of time. In clinical settings, there is not much time, plus healthcare professionals do not always have the time to see the children on a regular basis. Experts revealed that available measurement tools take up too much time. According to the literature, the CP-QoL CHILD takes 15-25 minutes to administer. Nevertheless, in the field, it appears to require more time due to additional explanations to the parents. When administering proxy reports, parents frequently need assistance, either because items are unclear to them or due to illiteracy reasons additional guidance is needed. Generally, experts acknowledged that the CP-QoL CHILD adequately addresses the essential domains for children with CP. Nevertheless, they did raise a concern that the grading system could be challenging for parents who are illiterate. The most notable aspect of the Peds-QL was its lack of focus on the child's well-being. It is focused on the limitations of the child. Lastly, the measurement tool brought up by the experts themselves was the PEDI. They expressed a favourable opinion towards it because it provides a rapid overview of the child's needs. Table 4 shows an overview of strengths and weaknesses of the existing tools according to experts.

CP-QoL CHILD	Strengths	Weaknesses
	<p>X10: "So I mean, even reading through the CP-QoL CHILD and KIDSCREEN, I like this. This could give you so much good information about where to target your rehab, not just giving a score. So I think that would be a strength."</p> <p>X10: "I think the one thing that stood out for me the CP-QoL CHILD. Was asking a parent about how they think their child feels about something."</p>	<p>X4: "Firstly, even looking at how it's graded. It's very hard on the ground, in and on the low resource society, to where parents may have limited education to really for them to understand certain markers, such as very unhappy and happy. And then, even worse, when it's graded with numbers it can be very confusing for parents."</p>
	<p>X8: "The CP-QoL CHILD is good as a starting point to see where the child's at."</p>	<p>X8: "It's quite orientated towards the west and I don't think it would be especially good for middle income countries."</p>
	<p>X4: "Oh, I think in general, I do like the questionnaire, and because it covers the key areas that are important in CP, it is covering communication, it is covering health and it is covering participation. And all those things are important. So I think generally I do like the questionnaire."</p>	<p>X9: "...when you look at the scales which: "have never" "they have almost", "never been" "sometimes" and "often" like "almost always" though, if translated, there is a confusion around "often: and "almost always" between. When you're administering it to the parents. They don't seem to understand. What's the difference between "often" and "almost always". So we try to break it down. But sometimes it doesn't come out easily to them."</p>
		<p>X8: "There are too many options, it goes 1 to 9 the scale is too long, and some of them overlap more than one box. That's what I don't like about that one."</p>
KID- SCREEN	<p>X8: "I like the way the KIDSCREEN is set out. So it's really clear about. "Have you been in a good mood?" "Never, seldom, quite often, very often always." I think you can you would easily choose one of those categories."</p>	
	<p>X9: "It's the KIDSCREEN and PEDS-QL to a general. They can be used for all types of disabilities."</p>	
PEDS- QL	<p>X9: "...in our setting we have used PEDS-QL in particular, which is a tool which has a different domain that captures physical, emotional, worry. Their activities family relationship and</p>	<p>X9: "I feel it's not conclusive, because if they don't capture anything around the child's well-being. It's more limited to limitations that the child can't do which</p>

<p><i>communication with the children. But I don't know how it's summarized, because we always use the proxy reports we tend to deal with mothers who are very lethal, sorry children who are very lethal so they cannot self answer this questionnaires. Yes, so it's usually the mother giving us the feedback for all these. That's the tool I personally used, and I feel it's good."</i></p>	<p><i>problems they are facing you, they don't capture the positive aspects..</i></p> <p><i>.. it's more into the negatives, it's more into that negative. So it's kinda limits."</i></p>
<p>PEDI</p> <p><i>X3: "Then I like the PEDI best, because it's giving me a also, because you can't see the children that often. So you have to make a complete difference... So they are more helped with for example aid like a sitting support, or a bar, or whatever they can stand on. Then on daily 2 or 3 times a day exercise."</i></p>	

Table 4 overview strengths and weaknesses according to experts.

The Paediatric Evaluation of Disability Inventory (PEDI) which was named lastly in Table 5 is developed to measure functional status, for children from 6 months to 7,5 years. Functional status by the authors referred to the extent to which a child is independent of his parents in performing daily activities.⁵⁸ While functional status may influence QoL they are not interchangeable terms. Therefore, functional status cannot be used as the sole indicator to measure QoL.²¹

Finally, the experts discussed challenges that they faced when using existing measurement tools. As mentioned before, time poses a barrier to the use of measurement tools. In settings where time is limited, experts often chose not to make use of existing tools. Expert X8 highlighted that sometimes parents do not fill in the answers they want to select but rather fill in the things that they think you (as a healthcare professional) want to hear. Expert X10 provided insights into existing mistrust towards 'foreigners' or 'outsiders' who approach individuals to fill in paperwork. Furthermore, X10 emphasized that the term "independence," as exemplified in the CP-QoL CHILD, is a concept predominantly rooted in Western culture. In contrast, in various other cultural contexts, the significance of such a concept might not be as pronounced, as these cultures uphold different norms and values. Another point discussed by X7 was the ability of children to fill in the forms. X7 shared:

X7: "I remember that it was quite a difficult aspect for the children in electric wheelchair they also answered questions. And there was one of the questions: do you like playing soccer? Yes, I like playing soccer, and the question was how satisfied are you in playing soccer? Well, they filled in they were very satisfied. But they were in the electric wheelchair. That's very difficult for interpretation such aspects, it's good that they feel well about it. But it's not a realistic approach for them. Are able to do you understand the problem?."

5.5 Experts' insights on tool usage for CP children

Experts' recommendations included the utilization of a measurement tool with context specific details as well as the involvement of children and their parents/caregivers. Asking

parents and caregivers what the child needs for improving their QoL seems crucial. While most western trained healthcare professionals have specific therapy goals and interventions in mind, it is important to get parents' and caregivers' input. The input of parents and caregivers is indispensable for a more comprehensive understanding of QoL. The interconnection is so strong that experts stated that if the family is doing well the child is doing well. The overall recommendation was to have proxy reports for the parents and self-reports for the children who are able to self-report. Asking children separately is essential because they might give you different information when parents and caregivers are around. The following text shows a part of the interview between researcher BS and expert X1:

BS: "What is in your opinion the best way to measure quality of life in children with CP living in low- and middle- income countries."

X1: "For me, it would be to ask the parents and the guardians."

BS: "Yeah?"

X1: "How can, what would they think would be a better quality of life for their children."

BS: "Okay."

X1: "Because what would make it easier for the child and what would make it easier for the guardian."

Experts X5 responding to the question of whether to include the child or the parents in a QoL measurement tool:

X5: "Yeah, it depends on the age of the child."

BS: "If the child is old enough?"

X5: "Then we are gonna ask both. Yeah. Separate,, not when they are together, then you get also a nice information sometimes."

BS: "You do this on purpose separately?"

X5: "Yeah, because the parents, some sometimes have other goals than the child. You have to work with both."

Another recommendation that was made by two experts was to make use of existing tools, despite the challenges it might pose. The advantage of doing so lies in the fact that existing measurement tools are supported and validated by literature. Additionally, most of the experts found it to be important to have a questionnaire simple and easy to administer. In order to use all resources it should be feasible for community health care workers and all other formal and informal health staff to administer the questionnaire. All parents and caregivers should be able to fill in the questionnaire regardless of their educational background. For example, a questionnaire that uses pictures or smileys instead of Likert scales would facilitate the use for illiterate people. As per expert opinions:

X6: "I think the best way to measure it.. is to make the measurement simple and short. Because considering the complexity of the quality of life as I said, considering individual functional level for the child and looking at the community aspect, and that the society or political for this level aspect, having this kind of all elements in the measurements, but try to keep it simple and default and more practical."

X10: "It is good to when you, quality of life that you can measure the participation of the ICF and not only the, the auto domains, like the function and structures and the activities, but also the participation part. And that's most of the time with questionnaires, I think. Yeah. Um, so that's still important. I think that is focused on that... I think it should, it should be easy for everyone, as we say before like community workers."

6) Discussion

This qualitative study aimed to examine the applicability and appropriateness of existing measurement tools that evaluate QoL for children with CP in LMICs. The results of this study reveal that the definition of QoL is not universally applicable. Additional QoL domains were identified, domains that differ from those known from conventional QoL literature. Domains included in existing QoL measurement tools should be considered carefully, taking cultural and context-specific details into consideration. Among existing measurement tools, the CP-QoL CHILD seems to be the strongest measurement tool available to measure QoL in children with CP. Despite this, experts strongly advise exercising special attention and consideration when using existing measurement tools that are developed in HICs and administering them in LMICs: the CP-QoL CHILD being such a tool.

6.1 Domains

While the literature lacks specific information about QoL domains for children with CP living in LMICs, studies conducted in HICs have identified important QoL domains for such children. The following section will present an overlap between QoL domains found in the literature of a study done in HICs and those that emerged from this study i.e. from interviews with experts. From the qualitative interviews conducted with families registered in the Victorian Cerebral Palsy Register at the Royal Children's Hospital in Melbourne, Waters et al. (2004) extracted thirteen distinct themes.²⁵ These themes laid the base for the development of the CP-QoL CHILD.⁴⁸ The identified domains encompass: physical health, body pain and discomfort, daily living tasks, participation in regular physical and social activities, emotional well-being & self-esteem, interaction with the community, communication, family health, supportive physical environment, future QoL, provision of & access to services, financial stability and social well-being.²⁵ The following section illustrates the alignments and/or discrepancies between the domains identified by the experts and the established domains from the study conducted by Waters et al. (2004).

'Beliefs & Stigma' and 'Interaction with the community'

There is an interconnectedness between the interaction of the child with CP and their parents with the community and the domain Beliefs & Stigma. Experts shared that the attitudes and beliefs held by individuals in the community about CP can deeply impact how children with CP are perceived and treated. There are still misconceptions about the origin of CP, which is in line with the literature found on the causes of CP.⁵⁹ These misconceptions can lead to the exclusion of children with CP and their families. For instance, if there is a misunderstanding that CP is contagious or caused by witchcraft their interaction with the community can be influenced negatively. Social acceptance, being a valued member of the community, and being treated 'normally' are components of the domain 'Interaction with the community'. This closely relates to the domain Beliefs & Stigma' that emerged from the interviews with experts in this study. In summary, the interaction of children with CP and the community they live in is influenced by the beliefs people have towards CP.

'Family' and 'Family health'

The domain 'Family health' is part of the larger domain 'Family'. Experts shared that the well-being of the family is an important component of the child's QoL. This is also highlighted in the domain 'Family health'. Family health refers to: "*good parental emotional health, good family relations, and few restrictions on the family to go out socially.*"²⁵ Good parental emotional health is essential, as parents' emotional health can impact their ability to provide care and support to the child with CP. Having good family relations was also mentioned as important by one of the experts. Experts' insights revealed that in most African

cultures it is crucial to establish good family relations. In these cultures, strong family ties and supportive relationships are often central to social structures. A supportive and positive family environment can foster the child's sense of belonging and encourage their active involvement. Unlike previously discussed alignments, the final component of 'Family health': *few restrictions on the family to go out socially*, contradicts the insights provided by the experts. Due to prevailing stigmatization, families often refrain from participating in social outings. An expert conveyed that children with disabilities are frequently not included in those activities.

'Resources & Access' and 'Provision of and access to services'

For children with CP, a pivotal aspect is accessibility to quality healthcare and rehabilitation services. 'Resources & access' recognizes that having easy access to healthcare is crucial for individuals to maintain good health and to promote well-being. This aligns with the domain 'Provision of and access to services' because that refers to: *"having access to therapy, respite care, and having the support required."*²⁵ Both domains emphasize the need for adequate resources and access to improve the QoL of children with CP living in LMICs.

'Education' and 'Participation in regular physical and social activities'

'Education' is part of the larger domain 'Participation in regular physical and social activities' because this domain refers to: *"participating in school activities, sporting activities and community activities."*²⁵ This domain further underscores the importance of children attending school, aligning with the emphasis expressed by the experts.

'Food & Malnutrition' and 'Daily living task'

The domain of 'Food & Malnutrition' falls under the broader category of 'Daily Living Tasks,' including: *"the ability to carry out normal daily living tasks including dressing, feeding and toileting and being independent."*²⁵ Particularly, the aspect of feeding is integral to this domain, as it directly corresponds to the domain of 'Food & Malnutrition.' Feeding is a fundamental activity of daily living and essential for maintaining well-being. One of the experts also raised the topic of toileting. Interestingly, this aspect was discussed within the domain 'Education'. For the reason that independent toileting was crucial for children to attend school. Despite not directly fitting into the 'Food & Malnutrition' domain, it was still highlighted by one of the experts. One of the experts also touched upon the subcategory "independence" but the discussion took a different direction. The expert's perspective on independence was characterized as having a strong Western influence, as notions of individual independence align more with Western values. The concept of independence may not hold the same relevance across all cultures, especially within LMICs.

6.2 CP-QoL CHILD

The CP-QoL CHILD measurement tool emerged as the most robust and potent QoL assessment tool currently available. This section intends to reflect on the applicability and appropriateness of the questionnaire in LMICs based on experts' insights. It will focus on the chapters encompassed within the CP-QoL CHILD proxy parent report 4-12 years. The chapters have been developed based on the inclusion of seven domains derived from the literature discussed in the preceding section of this discussion. The chapters of the CP-QoL CHILD will be guiding this section. The chapters are family and friends, participation, communication, health, special equipment, pain and bother, final questions, access to services, and your health.⁴⁹

'Family and friends'

This chapter emphasizes the importance of having good relationships with family and friends. Questions concern the way the child feels about how they get along with people in general, brothers, sisters, and other adults. This aligns with experts' opinions who also stated that having good relationships is indispensable for a good QoL. The question: "*the way they get along with their teachers and/or carers?*" indirectly implies that children are enrolled in school or that they have various carers. Both the experts from this study and the existing literature underscore the fact that in many LMICs, children with CP are often not enrolled in schools.¹ While this questionnaire lacks a separate chapter focused solely on school and education, many of the existing chapters include questions that refer to educational aspects. However, if children are not attending school, these questions can be disregarded, which accounts for the majority of CP children.

'Participation'

The category 'Participation' incorporates questions about children's ability to participate in school. In spite of that, according to experts, it is not comprehensive enough to capture all that is important for this category. For instance, the CP-QoL CHILD lacks the inclusion of crucial factors such as access to education, facilitation of infrastructure, and social interactions at school, which are specific challenges faced by children living LMICs. It seems that these important aspects are not adequately addressed in the CP-QoL CHILD tool. Including such subdomains helps to assess whether children with CP have equal opportunities as their typically developing peers.

'Communication'

This chapter consists of three questions the way the child feels about communicating with people they know, people they don't know and the way other people communicate with them. The importance of communication for children's QoL was highlighted by only two experts. Even though experts did not extensively elaborate on it communication is a fundamental aspect of a child's interactions and daily life, influencing their overall well-being.

'Health'

Within the 'Health' chapter, there is an interweaving of questions that address the child's self-esteem one of the domains established by literature.^{25 55} Examples are: "*How does your child feels about the way they look*" and "*How does your child feels about themselves*". Self-esteem is not explicitly mentioned by experts. Nevertheless, it is linked with the domain Beliefs & Stigma emerged by experts. While the questions about self-esteem are important it might not fully capture the broader concept of beliefs and stigmatization and their impact on a child's QoL. It therefore might not fully capture the complexities and wide-ranging effects of negative beliefs and stigmatization on a child's life. Furthermore, it encompasses just one question regarding the child's ability to drink independently, completely overlooking inquiries about feeding. This discrepancy contradicts the insights provided by experts, who emphasized the common challenges associated with feeding children with CP.

'Special equipment'

The 'Special equipment' chapter encompasses three questions. Despite experts underscoring the importance of special equipment for enhancing the QoL of children with CP, access to such equipment remains limited in many LMICs. One expert shared that she often encounters blank responses in this section.

‘Pain and bother’

The following four questions are included in this chapter “*Is your child bothered by hospital visits?*”, “*Is your child bothered when they miss school for health reasons?*”, “*Is your child bothered by being handled by other people?*” and “*Does your child worry about who will take care of them in the future?*” Half of the questions relate to school or the availability of resources. If the child is neither attending school nor has access to a hospital, half of this chapter will be unanswered. However, the majority of the experts agreed upon the fact that pain is an important aspect of the QoL of a child with CP and should be included in a measurement tool.

‘Final questions’

This chapter highlights different aspects of QoL for children with CP. Questions that are being asked correspond with the insights experts have shared.

‘Access to services’

A strong aspect of the CP-QoL CHILD is the chapter ‘Access to services’ which holds significant importance, particularly in LMICs because of severe shortages of rehabilitation workers. It correlates with the domain “Resources & Access” emerged by the interviewed experts in this study.

‘Your health’

The inclusion of questions concerning the health of the caregiver in the proxy report is highly valuable. For the reason that experts pointed out that a well-functioning family positively impacts the child’s well-being.

‘Expanding on existing chapters’

A noteworthy aspect to emphasize is that the CP-QoL CHILD does not encompass child safety. Waters et al. (2004) revealed that parents mentioned that child safety is important to them.⁵⁵ An expert interviewed in this study also raised the same concern, providing an example of child abuse. A study conducted by Power et al. (2018) additionally indicates that children with disabilities face a higher risk of abuse.¹⁵ Consequently, this domain warrants consideration for the future development of questionnaires concerning children with CP in LMICs

Fostering applicability and appropriateness

According to the Australian Academy of Cerebral Palsy and Developmental Medicine, the CP-QoL CHILD is translated into 20 different languages: Arabic, Bahasa Indonesian, Bahasa Malaysian, Dutch, Farsi, French, German, Greek, Hebrew, Italian, Korean, Mandarin, Myanmar, Polish, Portuguese, Serbian, Spanish, Tamil, Thai, and Turkish. Some of the languages are associated with countries classified as LMICs. The tool is translated into different languages with the guidance of the CP-QoL Translation Manual.⁶⁰ This consists of six steps: forward translation, reconciliation of items, backward translation, review of the forwards & backward translation, pre-test, and validation study. The process is comprehensive and takes multiple important steps into account. The second step, the reconciliation of items is key to ensure its appropriateness in the target culture.⁶¹ It includes focusing on differences in culture and linguistics that can cause barriers when adapting the English version to the target language.⁶⁰ Nevertheless, this process does not contain investigating the appropriateness of the construct being measured. According to Skevington et al. (2004) only translating the items is not efficient enough.⁶¹ This ethnocentric approach will have its limitations in LMICs. An ethnocentric strategy assumes that conceptual

dimensions of health identified by the measurement tool are transferable to the target culture.⁶¹ In order to assess the tool for full applicability to the target culture, a full cross-cultural adaptation or validation is needed. The CP-QoL CHILD is only culturally adapted in the Portuguese language for the use in Brazil and cross-culturally validated in Nepali.^{62 63}

6.3 Limitations and strengths

In order to enhance the validity and rigor of the qualitative analysis, it would have been optimal for the data coding to be carried out by two or more researchers independently. However, in this case, the coding was done by a single researcher (BS). This might have introduced bias because the interpretation and categorization of the data relied solely on one perspective.⁶⁴ Another bias that might have occurred is selection bias. It should be noted that the selected experts all work in the field of paediatric disabilities, this may indicate a strong interest in the topic. Consequently, this may have led to influenced results due to selection bias.⁶⁵ The expert panel had a wide variety of backgrounds with the majority being physiotherapists, which possibly biased the results as well. The researcher was aiming for data saturation, but this was not achieved due to time constraints. This may have led to compromised content validity.⁶⁶

The phenomenological nature of this research allowed the researcher to explore the perceptions of experts on the use of existing QoL measurement tools in CP children residing in LMICs. The topic guide that was used during the interviews was developed based on a literature review. There is a possibility that important themes have been missed because of the limited literature that is available in relation to the research topic. Literature about the appropriateness and applicability of existing disability measures is scarce.⁶⁷ In the literature the CP-QoL CHILD is seen as the strongest QoL measurement for CP children, for this reason, the topic guide consisted of questions about the CP-QOL CHILD. This could have resulted in the CP-QoL CHILD being the measurement tool named most frequently.

By excluding generic measurement tools, possible eligible tools may have been missed. The WHOQOL-100 is the most widely used QoL measurement tool in the world.⁶⁷ The development of the WHOQOL-100 included 15 international field centers, including LMICs.⁶⁸ With the aim to use the measurement tool in different countries and cultures. WHOQOL-100 consists of 100 questions, a shorter version is available namely WHOQOL-Bref which seems more suitable for routine use. However, the WHOQOL-Bref has an age limit, and its use for children is not validated. The WHOQOL group recognizes the importance of children's WHOQOL, this work has started in Thailand.⁶⁷ Another widely used QoL instrument is the KIDSCREEN, a generic QoL measurement tool that contains 10 life domains however not included in this study. Davis et al. (2008) argue that a generic instrument such as KIDSCREEN might not be specific enough to capture the unique challenges CP children face.⁶⁹ Domains important to them are not included in the tool which may cause higher QoL scores on the KISCREEN.⁶⁹ Another tool that was not discussed in this study is the Standard Capability Assessment (SCA). This tool was not found during the initial search but was brought to attention by one of the experts. The SCA was developed as a healthcare assessment for children with disabilities and is culturally appropriate for developing countries. This means that this tool holds good potential to measure the QoL for children with CP.⁶⁷

One limitation of this study is that it solely focuses on the applicability and appropriateness of QoL measurement tools. There are more psychometric properties that are important to take into consideration when choosing the right measurement tool. For the use of clinical practice,

the literature describes considering brevity, scoring, interpretability, and ease of administration.⁷⁰ Even though these properties fell out of the scope of this study, some experts commented on some of the aspects.

The literature research was compromised to google scholar which might have resulted in limited outcomes. Other search engines such as PubMed, MEDLINE and CINAHL are not being used in this study. The limitation to English language only could have resulted in missed articles. Nevertheless, a systematic review that has been done by Solans et al. (2007) showed that disease-specific QoL instruments were predominantly developed in English-speaking countries for example the United States, United Kingdom, and Canada.⁷⁰ This diminishes the change of missed articles.

7) Conclusion and recommendations

The research findings suggest that the existing measurement tools developed in HICs may not be fully appropriate nor applicable to the diverse contexts of LMICs. As per experts, the definition of QoL varies between children with CP living in HICs to children with CP in LMICs. The nuances in the definition of QoL possibly arise from diverse cultural, social, and contextual factors that influence the lives of children with CP. Certain questions that are presented in existing measurement tools may not hold relevance in LMICs. Existing QoL measurement tools are typically developed in collaboration with children and caregivers from HICs. However, to ensure a comprehensive understanding of all important aspects of QoL for children in LMICs, it is necessary to take into account cultural and context-specific details.

Important QoL domains derived from literature done in HICs show an overlap with those highlighted by experts. This research identified extra domains, not described in the literature nor captured by existing measurement tools, that are important to children with CP in LMICs. Beliefs around CP and stigma seem to play a pivotal role in the QoL of children with CP. Existing questionnaires do not put enough focus on these domains considering the importance of it.

Notwithstanding the challenges in cross-cultural applicability, the CP-QoL CHILD stands out as the strongest disease specific measurement tool currently available for assessing QoL in children with CP. Its comprehensive nature and focus on CP-related aspects have proven valuable. Regrettably, its applicability in LMICs remains uncertain because of its time-consuming nature. The administration of a questionnaire should not take longer than 15-20 minutes. Despite the fact that the literature states that the administering time is under 20 minutes, field experts share different experiences. In addition, the appropriateness can be questioned for the reason that some questions are not relevant for LMICs. In conclusion, given the limitations of existing measurement tools, CPA should pay careful attention when using tools developed in different contexts.

Recommendations for CPA:

Until a new tool is developed, the use of CP-QoL CHILD seems to be the most suitable for measuring the QoL of children with CP in LMICs. Experts advice on the use of existing tools because of the support provided by literature. Existing measurement tools have been tested and psychometric properties have been established. Yet, the sensitivity of the CP-QoL CHILD is not been established. In addition to the CP-QoL CHILD, it is recommended to use other assessment tools or indicators that are widely accepted and validated in LMICs to provide a more comprehensive evaluation of QoL in children with CP. Despite the fact that generic measurement tools have been excluded from this study the WHOQOL BREF has succeeded in developing a culturally appropriate measurement tool. When the additional version for children will be finalized this can be a potential strong QoL measurement tool. It is worth noting that the SCA also represents another promising measurement tool that merits exploration in the future. Its potential as a robust assessment method for evaluating the QoL in children with CP in LMICs seems evident.

Recommendations for a future QoL measurement tool for children with CP living in LMICs: First, focus group discussions with parents/caregivers and children from LMICs should be conducted to gain deeper insights into the perception of QoL. With the goal to attain a more profound insight into the unique challenges children with CP in LMICs face. Second, a tool should include the well-being of the family. Exclusively looking at the QoL of the child does not capture the broader scope. Third, the standard of 15 minutes administering time should be taken into account. Experts shared that time is valuable and scarce in the working field. In many instances, healthcare professionals or community healthcare workers do not have the time to administer a questionnaire. This is holding them back from using questionnaires in their practice. With the possible consequence that their interventions are not evaluated and ultimately not improved. Fourth, it is crucial to develop a tool that can be used by illiterate parents as well. In many LMICs, parents need assistance to fill in the questionnaire. This demands time and puts pressure on healthcare professionals. Fifth, incorporating questions about the beliefs and stigma will enhance the measurement of the QoL of children with CP. Sixth, including questions about school, helps to assess whether they have equal opportunities. It addresses important issues like enrolment, availability of special education services, and accessibility to school facilities. Lastly, questions about feeding the child should be included as well for a comprehensive questionnaire. In summary, future development of a QoL measurement tool should focus on:

- Understanding QoL for children with CP in LMICs
- Include family-well being
- Being time concise
- User-friendliness, for all users regardless of their level of literacy.
- Addressing beliefs and stigmatization
- Comprehensive inclusion of education
- Incorporating feeding strategies

Incorporating culturally sensitive and context-specific elements in the assessment of QoL for children with CP in LMICs seems indispensable. Future research in the field of QoL measurement for children with CP in LMICs should focus on developing a new tool that incorporates the unique challenges faced by these children. Collaborative efforts between field experts, parents, and CP children from LMICs can help establish a more applicable and culturally relevant measurement tool. Hopefully, this will contribute to a more holistic understanding of QoL and promote targeted interventions tailored for CP children in LMICs. This would ultimately improve the lives of children with CP in LMICs.

8) References

1. Jahan, I., Muhit, M., Hardianto, D. & Laryea, F. Epidemiology of cerebral palsy in low- and middle-income countries : preliminary findings from an international multi-centre cerebral palsy register. (2021).
2. Rosenbaum, P. *et al.* A report : the definition and classification of cerebral palsy. 8–14 (2006).
3. Pakula, A. T. & Braun, K. V. N. Cerebral Palsy : Classification and Epidemiology. 20, 425–452 (2009).
4. Gladstone, M. A review of the incidence and prevalence , types and aetiology of childhood cerebral palsy in resource-poor settings A review of the incidence and prevalence , types and aetiology of childhood cerebral palsy in resource-poor settings. (2014).
5. Olusanya, B. O. *et al.* Global prevalence of developmental disabilities in children and adolescents : A systematic umbrella review. (2023).
6. Donald, K. A., Samia, P., Kakooza-Mwesige, A. & Bearden, D. Pediatric cerebral palsy in Africa: A systematic review. *Semin Pediatr Neurol* 21, 30–35 (2014).
7. Bright, T., Felix, L., Kuper, H. & Polack, S. A systematic review of strategies to increase access to health services among children in low and middle income countries. *BMC Health Serv Res* 17, 1–19 (2017).
8. Chao, F., You, D., Pedersen, J., Hug, L. & Alkema, L. National and regional under-5 mortality rate by economic status for low-income and middle-income countries: a systematic assessment. *Lancet Glob Health* 6, e535–e547 (2018).
9. Eunson, P. Aetiology and epidemiology of cerebral palsy. *Paediatrics and Child Health (United Kingdom)* 22, 361–366 (2012).
10. Anttila, H., Autti-rämö, I., Suoranta, J., Mäkelä, M. & Malmivaara, A. Effectiveness of physical therapy interventions for children with cerebral palsy : A systematic review. 10, 1–10 (2008).
11. Mahmudul H., Imam A., Israt J., Muhit M., Hayley S., Badawi N., and Khandakerb G. Predictors of Rehabilitation Service Utilisation among Children with Cerebral Palsy (CP) in Low- and Middle-Income Countries (LMIC): Findings from the Global LMIC CP Register. *Brain Science*. (2005).
12. Larsson, I., Miller, M., Liljedahl, K. & Gard, G. Physiotherapists' experiences of physiotherapy interventions in scientific physiotherapy publications focusing on interventions for children with cerebral palsy: a qualitative phenomenographic approach. *BMC Pediatr* 12, 1 (2012).
13. King, S., Teplicky, R., King, G. & Rosenbaum, P. Family-Centered Service for Children with Cerebral Palsy and Their Families: A Review of the Literature. *Semin Pediatr Neurol* 11, 78–86 (2004).
14. World Health Organization & World Bank. (2011). World report on disability 2011. World Health Organization. *WORLD REPORT ON DISABILITY. ISBN 978 92 4 068521 5* <https://apps.who.int/iris/handle/10665/44575> (2011).

15. Power, R. *et al.* Health-related quality of life of children and adolescents with cerebral palsy in low- and middle-income countries: a systematic review. *Dev Med Child Neurol* 60, 469–479 (2018).
16. Brooks, R. & Konopad, E. Children's Quality of life measures. *Crit Care Med* 24, 1769 (1996).
17. Barcaccia, B. *et al.* Defining quality of life: A wild-goose chase? *Eur J Psychol* 9, 185–203 (2013).
18. Kaplan, R. M. & Ries, A. L. Quality of life: Concept and definition. *COPD: Journal of Chronic Obstructive Pulmonary Disease* 4, 263–271 (2007).
19. WHOQOL - Measuring Quality of Life| The World Health Organization. <https://www.who.int/tools/whoqol>. (2012).
20. Fayed, N. *et al.* Generic patient-reported outcomes in child health research: A review of conceptual content using World Health Organization definitions. *Dev Med Child Neurol* 54, 1085–1095 (2012).
21. Waters, E. *et al.* Quality of life instruments for children and adolescents with neurodisabilities: How to choose the appropriate instrument. *Dev Med Child Neurol* 51, 660–669 (2009).
22. Playford, D. The International Classification of Functioning, Disability, and Health. *Oxford Textbook of Neurorehabilitation* 3–7 (2015) doi:10.1093/med/9780199673711.003.0001.
23. Classification, I. *International Classification of Functioning , and Disability*. THE ICF: AN OVERVIEW. (2001).
24. Chen, K., Tseng, M., Shieh, J., Lu, L. & Huang, C. Research in Developmental Disabilities Determinants of quality of life in children with cerebral palsy : A comprehensive biopsychosocial approach. 35, 520–528 (2014).
25. Waters, E., Maher, E., Salmon, L., Reddihough, D. & Boyd, R. Development of a condition-specific measure of quality of life for children with cerebral palsy: Empirical thematic data reported by parents and children. *Child Care Health Dev* 31, 127–135 (2005).
26. Davis, E. Measuring the quality of life of children with cerebral palsy : comparing the conceptual differences and psychometric properties of three instruments. (2009).
27. Makris, T., Dorstyn, D. & Crettenden, A. Quality of life in children and adolescents with cerebral palsy: a systematic review with meta-analysis. *Disabil Rehabil* 43, 299–308 (2021).
28. Bjornson, K. F. & McLaughlin, J. F. The measurement of health-related quality of life (HRQL) in children with cerebral palsy. 8, 183–193 (2001).
29. Narayanan, U. G., Weir, S. & Fehlings, D. L. CPCHILD manual and interpretation. *Health (San Francisco)* 10–25 (2007).
30. Schneider, J. W., Gurucharri, L. M., Gutierrez, A. L. & Gaebler-Spira, D. J. Health-related quality of life and functional outcome measures for children with cerebral palsy. *Dev Med Child Neurol* 43, 601–608 (2001).
31. Davis, E., Reddihough, D., Murphy, N., Epstein, A., Reid, S.M., Whitehouse, A., Williams, K., Leonard, H. and Downs, J. Exploring quality of life of children with cerebral palsy and intellectual disability: What are the important domains of life? *Child Care Health Dev* 854–860. (2017).

32. Carlon, S. *et al.* A systematic review of the psychometric properties of Quality of Life measures for school aged children with cerebral palsy. *BMC Pediatr* 10, 81 (2010).
33. Herdman, U. R. M. *et al.* The European KIDSCREEN approach to measure quality of life and well-being in children : development , current application , and future advances. 791–803 (2014)
34. Kagawa-Singer, M., Padilla, G. V. & Ashing-Giwa, K. Health-Related Quality of Life and Culture. *Semin Oncol Nurs* 26, 59–67 (2010).
35. Eremenco, S. L., Cella, D. & Arnold, B. J. the Health Professions METHOD FOR THE STATUS QUESTIONNAIRES. *EVALUATION & THE HEALTH PROFESSIONS, Vol. 28 No. 2, June 2005 212-232 DOI: 10.1177/0163278705275342* 28, (2005).
36. Angi Stone-MacDonald, Ph.D. Gretchen Butera, Ph. D. Cultural Beliefs and Attitudes about Disability in East Africa. *Phys. Rev. E* 2002, 24 (2011).
37. Mabugu, T., Revill, P. & van den Berg, B. The methodological challenges for the estimation of quality of life in children for use in economic evaluation in low-income countries. *Value Health Reg Issues* 2, 231–239 (2013).
38. Rosenbaum, P. L., Livingston, M. H., Palisano, R. J., Galuppi, B. E. & Russell, D. J. Quality of life and health-related quality of life of adolescents with cerebral palsy. *Dev Med Child Neurol* 49, 516–521 (2007).
39. Albrecht, G. L. & Devlieger, P. J. The disability paradox: high quality of life against all odds. *Soc Sci Med* 48, 977–988 (1999).
40. Chen, K. L. *et al.* The Cerebral Palsy Quality of Life for Children (CP QOL-Child): Evidence of construct validity. *Res Dev Disabil* 34, 994–1000 (2013).
41. Salazar, L. F., Crosby, R. A. & DiClemente, R. J. *Research methods in health promotion. Research methods in health promotion.* (2015).
42. Norlyk, A. & Harder, I. What makes a phenomenological study phenomenological? An analysis of peer-reviewed empirical nursing studies. *Qual Health Res* 20, 420–431 (2010).
43. Guest, G., Namey, E. & Chen, M. A simple method to assess and report thematic saturation in qualitative research. *PLoS One* 15, 1–17 (2020).
44. Sappleton, N. & Lourenço, F. Email subject lines and response rates to invitations to participate in a web survey and a face-to-face interview: the sound of silence. *Int J Soc Res Methodol* 19, 611–622 (2016).
45. Clarke V, Braun V, H. N. T. analysis. Q. psychology: A. practical guide to research methods. 2015 J. 1;3:222-48. Thematic analysis. *Journal Positive Psychology* 1–5 (2015).
46. Tong, A., Sainsbury, P. & Craig, J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. 19, 349–357 (2007).
47. Resident, S., Medical, G. & Dungarpur, C. Assessment of Quality of Life in Children with Cerebral Palsy. *IOSR Journal of Dental and Medical Sciences (IOSR-JDMS)* 18, 45–49 (2019).
48. Dphil, E. W., Davis, E. & Stevenson, R. Psychometric properties of the quality of life questionnaire for children with CP. 49–55 (2007).

49. Davis E, Davern M, Waters E, Boyd R, Reddihough D, Mackinnon A, G. H. Cerebral Palsy Quality of Life Questionnaire for Adolescents (CP QOL-Teen) Manual. *Dev Med Child Neurol* 52, 24–25 (2013).
50. Varni, J. W. The PedsQL in pediatric cerebral palsy : reliability , validity , and sensitivity of the Generic Core Scales and Cerebral Palsy Module. 442–449 (2006).
51. Baars, R. M. *et al.* The European DISABKIDS project: Development of seven condition-specific modules to measure health related quality of life in children and adolescents. *Health Qual Life Outcomes* 3, 1–9 (2005).
52. Health-Related Quality of Life in Children and Adolescents with Cerebral Palsy: A Secondary Analysis of the DISABKIDS Questionnaire in the Field-Study Cerebral Palsy Subgroup. *Neuropediatrics*, 47(02), 097–106. (2016).
53. Narayanan, U. G. *et al.* Initial development and validation of the Caregiver Priorities and Child Health Index of Life with Disabilities (CPCHILD). *Dev Med Child Neurol* 48, 804–812 (2006).
54. Mueller-Godeffroy, E., Thyen, U. & Bullinger, M. Health-Related Quality of Life in Children and Adolescents with Cerebral Palsy: A Secondary Analysis of the DISABKIDS Questionnaire in the Field-Study Cerebral Palsy Subgroup. *Neuropediatrics* 47, 97–106 (2016).
55. Parkinson, K. N., Rice, H. & Young, B. Incorporating children’s and their parents’ perspectives into condition-specific quality-of-life instruments for children with cerebral palsy: A qualitative study. *Value in Health* 14, 705–711 (2011).
56. Waters E, Davis E, Boyd R, Reddihough D, Mackinnon A, Graham HK, Lo SK, Wolfe R, Stevenson R, Bjornson K, Blair E & Ravens-Sieberer U. *Cerebral Palsy Quality of Life Questionnaire for Children (CP QOL-Child) Manual*. Melbourne: University of Melbourne. (2013).
57. Dayanand, K. & Olivia E. Atherton¹, Jennifer L. Tackett², Emilio Ferrer¹, and R. W. R. Cross-Cultural Measurement Invariance of Adolescent Self- Report on the Pediatric Quality of Life Inventory™ 4.0. *Physiol Behav* 176, 139–148 (2018).
58. Wassenberg, J. E. *Pediatric evaluation of disability inventory (PEDI) : calibrating the Dutch version*. (2005).
59. Gilmore, L. & Wotherspoon, J. Perceptions of Cerebral Palsy in the Australian Community. *Intl J Disabil Dev Educ* (2021)
60. Davis E, Davern M, Waters E, Boyd R, Reddihough D, Mackinnon A, Graham HK. *Cerebral Palsy Quality of Life Questionnaire for Adolescents (CP QOL-Teen) Manual. Translation guidelines*. Melbourne: University of Melbourne. (2013).
61. Sartorius, N. & Kuyken, W. Translation of Health Status Instruments. in *Quality of Life Assessment: International Perspectives* 3–18. Springer Berlin Heidelber. (1994).
62. Maria Presumido Bracciali, L. *et al.* *Quality of life questionnaire for children with cerebral palsy: translation and cultural adaptation for Brazilian Portuguese Language Journal of Human Growth and Development* 2013; 23(2): 154-163 *QUESTIONÁRIO DE QUALIDADE DE VIDA DE CRIANÇAS COM PARALISIA CEREBRAL (CP QOL-CHILD): TRADUÇÃO E ADAPTAÇÃO PARA LÍNGUA PORTUGUESA QUALITY OF LIFE QUESTIONNAIRE FOR CHILDREN WITH CEREBRAL PALSY (CP QOL-CHILD): TRANSLATION AND CULTURAL*

- ADAPTATION FOR BRAZILIAN PORTUGUESE LANGUAGE. Journal of Human Growth and Development* vol. 23. (2013).
63. Adhikari, S. P. *Translation with Cross-cultural Adaptation of Cerebral Palsy Quality of Life Questionnaire for Children into Nepali and its Psychometric Properties.* (2020).
 64. O'Connor, C. & Joffe, H. Intercoder Reliability in Qualitative Research: Debates and Practical Guidelines. *Int J Qual Methods* 19, 1–13 (2020).
 65. COLLIER, D., & J. M. Insights and Pitfalls: Selection Bias in Qualitative Research. *Word Politics*, 49(1), 56-91. 56–91 (1996).
 66. Fusch, P. I. & Ness, L. R. Are we there yet? Data saturation in qualitative research. *Qualitative Report* 20, 1408–1416 (2015).
 67. Skevington, S. M. *et al.* Developing methods for assessing quality of life in different cultural settings - The history of the WHOQOL instruments. *Social Psychiatry and Psychiatric Epidemiology* vol. 39 1–8 Preprint at <https://doi.org/10.1007/s00127-004-0700-5> (2004).
 68. Programme on mental health : WHOQOL user manual. *DIVISION OF MENTAL HEALTH AND PREVENTION OF SUBSTANCE ABUSE WORLD HEALTH ORGANIZATION.* (2012).
 69. Davis, E. *et al.* Quality of life of children with CP: Condition-specific instrument and proxy reports. *Dev Med Child Neurol* 50, 167 (2008).
 70. Solans, M. *et al.* Health-related quality of life measurement in children and adolescents: A systematic review of generic and disease-specific instruments. *Value in Health* 11, 742–764 (2008).

9) Appendix

Appendix 1: Invitation email

Dear Participant,

I obtained your contact information through Huib Cornielje my thesis advisor. My name is Bobbi van Kesteren and I am a master student at KIT Royal Tropical Institute Amsterdam.

I am sending you this letter because I would like to invite you to participate in an interview schedule which is part of my qualitative research. This research focuses on the applicability and appropriateness of existing questionnaires for measuring the quality of life in children with cerebral palsy in low- and middle-income countries (LMICs). By better understanding the challenges of measuring quality of life in these contexts, hopefully interventions can be improved in the future and will result in better outcomes for children with cerebral palsy.

As an expert in the field of cerebral palsy, your insights and experiences can be helpful to gain a deeper understanding about the strengths and weaknesses of using existing questionnaires in LMICs. The Pediatric Quality of Life Inventory (PedsQL) and the Cerebral Palsy Quality of Life Questionnaire (CP QOL-CHILD) are examples of questionnaires that are – at times - being used in LMICs.

The interview will be conducted remotely via Zoom and will take approximately 45 minutes. Your participation will be strictly confidential, and your insights will only be used for the purpose of this research. Please find attached an informed consent to protect your rights and welfare.

I would be honoured to have you participate in this study and contribute your valuable expertise. If you are interested in participating, please let me know your availability, then we can schedule a time that works best for you.

Thank you for considering this request.

Sincerely,

Bobbi van Kesteren

Appendix 2: Informed Consent

Introduction

My name is Bobbi van Kesteren, I am a master student International Health at the KIT Royal Tropical Institute. I am conducting a study on the appropriateness and applicability of existing measurement tools to evaluate quality of life in children with cerebral palsy (CP) living in low- and middle- income countries (LMICs).

The aim of the study is to examine the appropriateness and applicability of existing measurement tools to evaluate quality of life in children with CP in LMICs.

For the reason that you are a health professional working with children with CP in LMICs, I would like to invite you to be a part of this study.

Informed consent form

If you agree to participate, I hope that the information which you provide will help to develop a better understanding on how to measure quality of life in children living with CP in LMICs. The study takes place between January - August 2023.

Procedures including confidentiality

If you agree to participate in this study, I will use an in-depth semi-structured online interview which will last maximally 45 min. The conversation will be recorded, if you agree, for analysis purposes only. I will ask your opinion about existing measurement tools to evaluate quality of life in children with CP, applicability and appropriateness in LMICs. You can express your honest opinion freely in this interview because all the data will be anonymous at all times. Only the team of researchers (master student and the thesis advisor) will have access to the data set. The recorded files will be deleted 6 months upon the completion of the study.

In publications, the findings will focus on the broader context that will derive from the interview, not on your particular answers, so that nobody can recognise the setting and your opinions.

Risk, discomforts and right to withdraw

After having agreed to participate in the interview, you are still free to refuse to answer any question that makes you uncomfortable and it will not have any consequences for you. You can also withdraw from this study at any time. And lastly, no risk of participation is expected.

Benefits

This study may not help you directly, but the results will help to inform the future development of measurement tools to evaluate quality of life in children with CP living in LMICs.

Sharing the results

After the study is completed, we will share the results in a workshops with stakeholders relevant to the master International Health, including student representatives and alumni. If the participant wishes to receive a copy of the final thesis, on request one can be send afterwards.

Consent and contact

Do you have any questions that you would like to ask?

Are there any things you would like me to explain again or say more about?
Do you agree to participate in the interview?

DECLARATION: TO BE SIGNED BY THE RESPONDENT

Agreement respondent

The purpose of the interview was explained to me and I agree to be interviewed
..... (name of person).

Signed

Date

WITNESS SIGNATURE

Signed

Date

If you have any questions or want to file a complaint about the research you may contact:

Contact information organization F.Maldonado@kit.nl	Contact for Ethics Committee s.alba@kit.nl
--	---

Appendix 3: Topic guide

Data collection tool guide, in depth semi structured interview

Section 1: Quality of life

Question 1

Can you please tell me your profession and how many years of experience you have in working with children with CP in LMICs?

Question 2

How would you define QoL of children with CP living in LMICs?

Question 3

Why do you think measuring QoL in children with CP living in LMIC is important?

Question 4

Has the meaning of QoL changed over the last years?

Question 5

How does CP affect the QoL of children living LMICs?

Question 6

What is in your opinion the best way to measure QoL in children with CP living in LMICs?

Section 2: Inventory existing measurement tools

Question 7

Can you tell me about what you know about the available measurement tools to evaluate QoL in children with CP living in LMICs?

Question 8

Do you know the difference for example between the KIDSCREEN a generic measurement tool and the CP-QoL CHILD a disease specific measurement tool?

Question 9

Do you use measurement tools to evaluate the effect of your interventions on QoL? If yes, which ones?

Question 10

How often do you measure QoL? Do you want to measure change or to get a general impression of the child's wellbeing? Can you explain more about this?

(Question 11)

(If not why do you not make use of QoL measurement tools?)

Section 3: Usability and appropriateness

Question 12

Can you name strengths and weaknesses of existing measurement tools? For example, regarding the domains they measure, regarding practical feasibility to administer.

Question 13

What is in your opinion the best tool available to evaluate quality of life in children with cerebral palsy living in LMICs, and why?

Question 14

Are the available measurement tools user friendly? What do you think about the time it takes to conduct the questionnaires?

Question 15

Do you think that existing measurement tools cover all the domains of QoL in children with CP living in LMIC?

Question 16

How well do the existing measurement tools capture the unique experiences and challenges faced by children with CP living in LMICs?

Question 17

Are there any cultural or contextual factors that may affect the validity or reliability of existing measurement tools for evaluating QoL in children with CP in LMICs?

*Section 4: Other***Question 18**

What are the differences of QoL in children with CP living in LMICs compared to HICs?

Question 19

Is QoL the best outcome to measure change in well-being of a child with CP? Or do you think we should look at other outcomes?

Question 20

What recommendations would you make for future research in this area, in terms of QoL measurement tools and evaluation of interventions used when dealing with children with CP living in LMICs?

Question 21

Are there any important themes you missed in this interview that you would like to elaborate on when it comes to measuring quality of life in children with CP in LMICs?

Appendix 4: Code Tree

Code	Code Group 1	Code Group 2	Code Group 3	Code Group 4
Additional comments & under 2 yrs				Recommendations
Additional comments & under 2 yrs: Recommendation (for future tools measure tiny changes)				
Additional comments & under 2 yrs: Recommendation (future QOL tool, to have a box for additional comments)				
Additional comments & under 2 yrs: Recommendation (future tool, to make a tool for parents for children under two)				
Additional comments & under 2 yrs: Recommendation (tool that start at the age of 0 but then for caregivers)				
Additional comments & under 2 yrs: Recommendations (ask to elaborate on answers)				
Beliefs & Stigma			Key domains	
Beliefs & Stigma: Beliefs acceptance belonging				
Beliefs & Stigma: Beliefs and acceptance				
Beliefs & Stigma: Beliefs CP (abandoned)				
Beliefs & Stigma: Beliefs CP (affects the QOL)				
Beliefs & Stigma: Beliefs CP (another example)				
Beliefs & Stigma: Beliefs CP (are negative)				
Beliefs & Stigma: Beliefs CP (attitudes towards children with disabilities)				
Beliefs & Stigma: Beliefs CP (attitudes towards the child)				
Beliefs & Stigma: Beliefs CP (beliefs of the community affects the QOL of the child)				
Beliefs & Stigma: Beliefs CP (children are being bullied at school)				
Beliefs & Stigma: Beliefs CP (children are being hidden)				
Beliefs & Stigma: Beliefs CP (children are not taking out)				
Beliefs & Stigma: Beliefs CP (cp is unknown in LMICS)				
Beliefs & Stigma: Beliefs CP (difference between HIC and LMICS)				

Beliefs & Stigma: Beliefs CP (doctors make the child normal again)				
Beliefs & Stigma: Beliefs CP (example of unsupportive family)				
Beliefs & Stigma: Beliefs CP (it improved by taking children with disabilities on the national football field)				
Beliefs & Stigma: Beliefs CP (lack of acceptance)				
Beliefs & Stigma: Beliefs CP (mis understanding and concept of CP)				
Beliefs & Stigma: Beliefs CP (misconceptions of cerebral palsy)				
Beliefs & Stigma: Beliefs CP (negative attitudes towards CP children)				
Beliefs & Stigma: Beliefs CP (neglecting)				
Beliefs & Stigma: Beliefs CP (positive example)				
Beliefs & Stigma: Beliefs CP (positive, children like to help each other)				
Beliefs & Stigma: Beliefs CP (positive, people are being nice)				
Beliefs & Stigma: Beliefs CP (shame and angry)				
Beliefs & Stigma: Beliefs CP (sin)				
Beliefs & Stigma: Beliefs CP (stigma)				
Beliefs & Stigma: Beliefs CP (stigmatizing)				
Beliefs & Stigma: Beliefs CP (teacher are refusing CP children)				
Beliefs & Stigma: Beliefs CP (the attitudes from the family towards the child with CP)				
Beliefs & Stigma: Beliefs CP (traditional healers pose stigma)				
Beliefs & Stigma: Beliefs CP (wanting their child to become normal)				
Beliefs & Stigma: Beliefs CP (we need to get rid of stigma by traditional healers)				
Beliefs & Stigma: Beliefs CP children (they don't want to show the child)				
Beliefs & Stigma: Yeah, and you see, that makes also difficult in a treatment for the ac				
Challenges		Existi ng tools		
Challenges: Challenge (mistrust)				
Challenges: Challenge in measuring QOL				
Challenges: Challenge measuring QOL in children with CP				
Challenges: Challenge measuring QOL in children with CP (cultural maybe)				
Challenges: Challenges with asking parents				
Challenges: Challenges in measuring QOL in children with cerebral palsy				

Challenges: Challenges in measuring QOL in children with CP				
Challenges: Challenges in measuring QOL in CP children				
Challenges: Time (But I cannot do this in my therapy session cannot do this in a consult)				
Challenges: Time (reason why not to use measurement tools)				
Challenges: Time (So you'd find that in the hospitals this would never be administered)				
Challenges: Time (why not used it)				
Challenges: Time as a barrier not to use tools				
Challenges: Time as a component why not to use tools				
Context specific				Recommendations
Context specific: Context specific (of the concept QOL)				
Context specific: It's about being together and honouring the being together. And that's				
Context specific: Recommendation (concept easy to transfer in less time)				
Context specific: Recommendation (continent specific)				
Context specific: Recommendation (country specific)				
Context specific: Recommendation (including participation)				
Context specific: Recommendation (interview)				
Context specific: Recommendation (not one tool for all)				
Context specific: Recommendation (start to as health professionals who work overseas)				
Context specific: Recommendation (The details need to be like, yeah country specific)				
Context specific: Recommendations (for future tools, domains to include)				
Context specific: Recommendations (for LMICS)				
Context specific: Recommendations (sit with the community)				
Education			Key domains	
Education: Education (difficulties for CP children)				
Education: Education (including school as part of rehab)				
Education: Education (majority don't go to school)				
Education: Education (part of QOL)				

Education: School (ability to go to school)				
Education: School (access to education)				
Education: School (CP children hardly go to school)				
Education: School (facilitate education for CP children)				
Education: School (importance of education)				
Education: School (importance of getting CP children in school)				
Education: School (in a CP child the brain is not always affected. They are clever)				
Education: School (is a focus in LMICS)				
Education: School (is an important domain to include)				
Education: School (is not accessible for CP children)				
Education: School (schools are not facilitating or helping CP children)				
Education: School (So if you just forget about learning, you forget about the thing he)				
Existing tools as a recommendation				Recommendat ions
Existing tools as a recommendation: Recommendation (use of an existing tool, supporting literature)				
Existing tools as a recommendation: Recommendation (using existing tool, and check for availability)				
Existing tools as a recommendation: Recommendations (for measuring QOL)				
Existing tools as a recommendation: Recommendations (using existing tools)				
Family			Key domai ns	Recommendat ions
Family: Involve parents and the whole family				
Family: Involve children				
Family: Involve family (as a recommendations for a tool)				
Family: Involve family (The family is doing well the child will do well)				
Family: Involve parent and children (but ask them separately)				
Family: Involve parents (involve mam, ask what she needs)				
Family: Involve parents (mothers and ask them what they need for the child)				
Family: Involve parents (QOL and family go together)				
Family: Involve parents (they see the problems)				

Family: Involve parents and children (and ask what they need)				
Family: Involve parents and children (but ask them separately)				
Family: Involve parents and children (they have different goals)				
Family: Involve parents and household for assessing QOL				
Family: Involving (So what does he think of his life? What would he like to do?)				
Family: Involving parent and family seeing it as one unit				
Family: Involving parents (and hear them)				
Family: Involving parents (and moms is important when their attitude is good towards the child, the well-being of the child will be better)				
Family: Involving parents (challenges that comes with administering questionnaires separately)				
Family: Involving parents (know what's important to them)				
Family: Involving parents (Moms and the family is crucial)				
Family: Involving parents (the importance of asking what they need because its very heavy)				
Family: Involving parents and children (ask them both and separately)				
Family: Involving parents and children (interview them)				
Family: Involving parents and children (to find out QOL)				
Family: Involving parents and family (ask what they need)				
Family: Involving parents and family example of its importance				
Family: Involving parents and guardians to improve QOL				
Family: Involving parents/guardians (best way to go)				
Family: Recommendation (tools that captures also family and child)				
Family: Recommendations (again a tool that captures the family)				
Food and malnourishment			Key domains	
Food and malnourishment: Food & Nutrition recommendation (Also a part about feeding problems)				
Food and malnourishment: Food & Nutrition				

Food and malnourishment: Food & Nutrition (a lot of malnourishment)				
Food and malnourishment: Food & Nutrition (challenge)				
Food and malnourishment: Food & Nutrition (importance of feeding)				
Food and malnourishment: Food & Nutrition (make it easier for them)				
Food and malnourishment: Food & Nutrition (mal nutrition is a big problem)				
Food and malnourishment: Food & Nutrition (malnutrition is a challenge)				
Food and malnourishment: Food & Nutrition (problems)				
Keep it simple				Recommendat ions
Keep it simple: Recommendation (for a future QOL tool)				
Keep it simple: Recommendation (future tools work with smiley faces)				
Keep it simple: Recommendation (include both subjective and objective measures)				
Keep it simple: Recommendation (tool for illiterate parents)				
Keep it simple: Recommendations (future tool)				
Keep it simple: Recommendations (future tools short be short and simple)				
Keep it simple: Recommendations (grading for parents)				
Keep it simple: Recommendations (short and not too heavy)				
Keep it simple: Recommendations (short and simple not time consuming)				
QOL definition	Conceptuali zing QOL			
QOL definition: Children (domains)				
QOL definition: Children (if you organize school)				
QOL definition: Children (important domains)				
QOL definition: Children (important aspects)				
QOL definition: Children (important domains for them)				
QOL definition: Children (important domains)				
QOL definition: Defining QOL (including all domains)				
QOL definition: Defining QOL (not without function)				
QOL definition: Defining QOL (pain is an important domain)				
QOL definition: Individualized (repeats one sits does not fit all)				

QOL definition: QOL (again individual)				
QOL definition: QOL (Defining QOL)				
QOL definition: QOL (meaning QOL for CP children in LMICs)				
QOL definition: QOL defining (asking them)				
QOL definition: QOL defining (big concept)				
QOL definition: QOL defining (cultural and context specific)				
QOL definition: QOL defining (cultural)				
QOL definition: QOL defining (culture and context specific)				
QOL definition: QOL defining (different domains)				
QOL definition: QOL defining (different levels different domains)				
QOL definition: QOL defining (different meanings)				
QOL definition: QOL defining (different to every child)				
QOL definition: QOL defining (does not have a correlation with functional status)				
QOL definition: QOL defining (domains that should be included)				
QOL definition: QOL defining (highly individualized)				
QOL definition: QOL defining (holistic and broad)				
QOL definition: QOL defining (holistic ICF)				
QOL definition: QOL defining (holistic)				
QOL definition: QOL defining (HRQOL is not QOL)				
QOL definition: QOL defining (important domains for CP children in LMICS)				
QOL definition: QOL defining (inconsistence)				
QOL definition: QOL defining (individualized)				
QOL definition: QOL defining (is individual and country specific)				
QOL definition: QOL defining (key aspects of QOL)				
QOL definition: QOL defining (measurements and the construct they measure)				
QOL definition: QOL defining (multiple domains)				
QOL definition: QOL defining (poor for children with CP)				
QOL definition: QOL defining (subjective)				
QOL definition: QOL defining (whole picture broad perspective)				
QOL definition: QOL defining it (well-being of the family)				

QOL definition: QOL definition (for children in CP children in LMICs)				
QOL definition: QOL definition (value laden)				
QOL definition: QOL definition also common experiences				
QOL definition: QOL importance (bigger picture)				
QOL definition: QOL is individual (no one size fits all)				
QOL importance	Conceptualizing QOL			
QOL importance: QOL (importance)				
QOL importance: QOL defining (important domains to include)				
QOL importance: QOL importance (example)				
QOL importance: QOL importance (for funding)				
QOL importance: QOL importance (good outcome)				
QOL importance: QOL importance (good way to measure well-being)				
QOL importance: QOL importance (important to use as an outcome measure)				
QOL importance: QOL importance (Of course. Yeah, yeah)				
QOL importance: QOL importance (the reason why measuring QOL is important)				
QOL importance: QOL importance (the way to go)				
QOL importance: QOL importance (to see the effect of your intervention)				
QOL importance: QOL importance (to see the effects of interventions)				
QOL importance: QOL importance (very)				
QOL importance: QOL importance (Yes, very important)				
QOL importance: QOL importance domain is engagement, sense of belonging.				
Resources & Access			Key domains	
Resources & Access: Resources (less physios compared to children who need therapy)				
Resources & Access: Resources & Access (access its a barrier)				
Resources & Access: Resources & Access (adaptive equipment)				
Resources & Access: Resources & Access (adaptive equipment) (2)				
Resources & Access: Resources & Access (adaptive equipment's)				

Resources & Access: Resources & Access (are different in LMICS)				
Resources & Access: Resources & Access (as the biggest difference of QOL between HIC and LMIC)				
Resources & Access: Resources & Access (assistive devices)				
Resources & Access: Resources & Access (big influence for children with CP in LMICS)				
Resources & Access: Resources & Access (difficult to access facilities)				
Resources & Access: Resources & Access (having the right aid)				
Resources & Access: Resources & Access (negatively influences QOL for CP children)				
Resources & Access: Resources & Access (no accessibility to services)				
Resources & Access: Resources & Access (no special actions has been taken to get CP to school)				
Resources & Access: Resources & Access (not the same as in HIC)				
Resources & Access: Resources & Access (poverty)				
Resources & Access: Resources & Access (the amount of care children receive is very little)				
Resources & Access: Resources & Access I think a severe CP child, in lower income countries, they have not ve				
Resources & Access: Resources & Access important for CP children and their QOL				
Resources & Access: Resources & Access (not getting proper care)				
Resources & Access: Resources & Access (big difference between HIC and LMIC)				
Strengths existing measurement tools		Existing tools		
Strengths existing measurement tools: Strengths Ladder of Life (It's simple all caregivers understand it and it is very fast)				
Strengths existing measurement tools: Strengths existing tool (because its flexible)				
Strengths existing measurement tools: Strengths Ladder of Life (every caregiver understands it)				
Strengths existing measurement tools: Strengths PEDI (it's about daily activities)				
Strengths existing measurement tools: Strengths PEDI existing tools (functioning questions)				

Strengths existing measurement tools: Strength CP-QOL CHILD (generally she likes the questionnaire covers important domains for CP children)				
Strengths existing measurement tools: Strength CP-QOL CHILD (gives an overview where the child's at)				
Strengths existing measurement tools: Strength CP-QOL CHILD (good as a starting point)				
Strengths existing measurement tools: Strength CP-QOL CHILD (having an idea where the child's at)				
Strengths existing measurement tools: Strength CP-QOL CHILD (I'm satisfied with the domains)				
Strengths existing measurement tools: Strength CP-QOL CHILD (questions about communication are good)				
Strengths existing measurement tools: Strength CP-QOL CHILD and KIDSCREEN gives a lot of information				
Strengths existing measurement tools: Strength existing measurement tools (KIDSCREEN and PEDS-QL) as general tools can be used for all disabilities, that's an advantage in LMICS.				
Strengths existing measurement tools: Strength existing tool (PEDI)				
Strengths existing measurement tools: Strength KIDSCREEN (clear set out)				
Strengths existing measurement tools: Strength KIDSCREEN (good lay out)				
Strengths existing measurement tools: Strength KIDSCREEN and PEDS-QL (general tools always usable)				
Strengths existing measurement tools: Strength Ladder of Life (even a little bit older children they understand it)				
Strengths existing measurement tools: Strength PEDI (Then I like the PEDI best, because it's giving me a also, because you)				
Strengths existing measurement tools: Strength PEDS-QL (activation domain)				
Strengths existing measurement tools: Strength PEDS-QL (feels good about it)				
Strengths existing measurement tools: Strength TA-QOL (easy to use)				
Strengths existing measurement tools: Strengths (Before that, I use just numbers or so a smiley or something like that)				
Strengths existing measurement tools: Strengths PEDS-QL (emotional domain)				

Using existing measurement tools in LMICS		Existing tools		
Using existing measurement tools in LMICS: Applicability (appropriateness)				
Using existing measurement tools in LMICS: Applicability (covering the domains for children in Europe)				
Using existing measurement tools in LMICS: Applicability (cross and cultural validation)				
Using existing measurement tools in LMICS: Applicability (cross cultural validation isn't enough)				
Using existing measurement tools in LMICS: Applicability (does not want to use CP-QOL CHILD in LMICS)				
Using existing measurement tools in LMICS: Applicability (inappropriate questions)				
Using existing measurement tools in LMICS: Applicability (not all questions of PEDI are relevant in LMICS)				
Using existing measurement tools in LMICS: Applicability (not something that would be preferred the use of existing tools in LMICS)				
Using existing measurement tools in LMICS: Applicability (questions are not relevant)				
Using existing measurement tools in LMICS: Applicability (there is overlap)				
Using existing measurement tools in LMICS: Applicability (tool should be context specific)				
Using existing measurement tools in LMICS: Applicability (translating is not enough)				
Using existing measurement tools in LMICS: Applicability (would not be appreciated)				
Using existing measurement tools in LMICS: Applicability CP-QOL CHILD (not always usable in very small children without diagnosis) (2)				
Using existing measurement tools in LMICS: Applicability (not always usable in these settings)				
Weakness' existing measurement tools		Existing tools		
Weakness' existing measurement tools: Existing measurement tools weakness' (too long)				
Weakness' existing measurement tools: Weakness' CP-QOL CHILD				
Weakness' existing measurement tools: Weakness' CP-QOL CHILD (again grading for certain people in society)				

Weakness' existing measurement tools: Weakness' CP-QOL CHILD (asking about money etc can be interpreted different because different way of communicating)				
Weakness' existing measurement tools: Weakness' CP-QOL CHILD (few questions that are sometimes hard to explain or not relevant)				
Weakness' existing measurement tools: Weakness' CP-QOL CHILD (grading is too difficult)				
Weakness' existing measurement tools: Weakness' CP-QOL CHILD (grading with numbers is hard for parents to understand)				
Weakness' existing measurement tools: Weakness' CP-QOL CHILD (I think it was designed for a more western setting)				
Weakness' existing measurement tools: Weakness' CP-QOL CHILD (missing domain?)				
Weakness' existing measurement tools: Weakness' CP-QOL CHILD (not always relevant questions)				
Weakness' existing measurement tools: Weakness' CP-QOL CHILD (not always usable in very small children without diagnosis)				
Weakness' existing measurement tools: Weakness' CP-QOL CHILD (oriented towards the west)				
Weakness' existing measurement tools: Weakness' CP-QOL CHILD (questions about nutrition are missing)				
Weakness' existing measurement tools: Weakness' CP-QOL CHILD (quite long)				
Weakness' existing measurement tools: Weakness' CP-QOL CHILD (reason why she is not using the tool)				
Weakness' existing measurement tools: Weakness' CP-QOL CHILD (scales)				
Weakness' existing measurement tools: Weakness' CP-QOL CHILD (specific for children with CP, LMIC delayed diagnosis)				
Weakness' existing measurement tools: Weakness' CP-QOL CHILD (takes up more time in LMICS settings)				
Weakness' existing measurement tools: Weakness' CP-QOL CHILD (time consuming because of explaining)				
Weakness' existing measurement tools: Weakness' CP-QOL CHILD (too late)				
Weakness' existing measurement tools: Weakness' CP-QOL CHILD (too long)				

Weakness' existing measurement tools: Weakness' CP-QOL CHILD (you need more space for explaining answers)				
Weakness' existing measurement tools: Weakness' CP-QOL CHILD big amount of the questions concern special equipment not realistic in LMICS)				
Weakness' existing measurement tools: Weakness' CP-QOL CHILD has a big amount of questions that is about school)				
Weakness' existing measurement tools: Weakness' CP-QOL CHILD loses its accuracy and maybe validity because parents don't always get the questions right				
Weakness' existing measurement tools: Weakness' CP-QOL CHILD not always usable in LMICs				
Weakness' existing measurement tools: Weakness' CP-QOL CHILD the way questions are formulated				
Weakness' existing measurement tools: Weakness' existing measurement tool (too short is also not working)				
Weakness' existing measurement tools: Weakness' existing measurement tools (no one that captures the full domains of QOL for children with CP in LMICS)				
Weakness' existing measurement tools: Weakness' existing tools (challenge with tools)				
Weakness' existing measurement tools: Weakness' existing tools (not measuring QOL but functional status)				
Weakness' existing measurement tools: Weakness' existing tools (relevance)				
Weakness' existing measurement tools: Weakness' existing tools (resource demanding)				
Weakness' existing measurement tools: Weakness' existing tools (there are not many)				
Weakness' existing measurement tools: Weakness' existing tools (translation)				
Weakness' existing measurement tools: Weakness' KIDSCREEN (Designed for Europe)				
Weakness' existing measurement tools: Weakness' KIDSCREEN (questions are not relevant) (2)				
Weakness' existing measurement tools: Weakness' KIDSCREEN (some questions are inappropriate)				
Weakness' existing measurement tools: Weakness' KIDSCREEN and CP-QOL CHILD was just me				

Weakness' existing measurement tools: Weakness' KIDSCREEN-10 (too short)				
Weakness' existing measurement tools: Weakness' PEDI (not a lot on phycological issues)				
Weakness' existing measurement tools: Weakness' PEDI (paid version)				
Weakness' existing measurement tools: Weakness' PEDS-QL (did not capture the whole)				
Weakness' existing measurement tools: Weakness' PEDS-QL (negative)				
Weakness' existing measurement tools: Weakness' PEDS-QL (not being a strong measurement, not measuring well-being)				
Weakness' existing measurement tools: Weakness' PEDS-QL (not measuring well-being)				
Weakness' existing measurement tools: Weakness' PEDS-QL (scales not always understandable for parents)				
Weakness' existing measurement tools: Weakness (illiterate hard to answer for parents)				
Weakness' existing measurement tools: Weakness CP-QOL CHILD is not applicable for all settings and parents				
Weakness' existing measurement tools: Weakness CP-QOL CHILD takes more time to administer because of additional explaining that needs to be done				
Weakness' existing measurement tools: Weakness of the CP-QOL CHILD (severe levels can't fill in relies on proxy)				

Appendix 5: Ethical Clearance



KIT Royal
Tropical
Institute

RESEARCH ETHICS COMMITTEE

Contact: Sandra Alba
s.alba@kit.nl

To: Bobbi van Kesteren
bobbivk@hotmail.com

Amsterdam, 19-5-2023

Subject Decision Research Ethics Committee regarding S-212

Dear Bobbi van Kesteren,

The Research Ethics Committee (REC) of the Royal Tropical Institute has reviewed your application for a waiver for your thesis research on that was originally submitted on 26 April 2023 (S-212). The objective of the study is to examine how the quality of life of children with cerebral palsy (CP) can be measured in low- and middle-income countries.

Your proposal has been exempted from full ethical review based on the following considerations:

- a) the participants will be involved in their professional capacity only; the issues to be covered in the topic list cover information related to the duties of the respondents and information in the public domain; questions related to any personal questions are not included;
- b) the participants will be asked informed consent before the data collection. This to make sure voluntary and informed participation is taking place and the participant can decide to decline or withdraw participation at any moment during the process without any effect on reputation, or other consequences;
- c) participating in this study does not bear any physical, psychological and/or socio-economical risk or discomfort;
- d) all information will be derived, processed, stored and published anonymously;
- e) the research has important social, educational or scientific value.

This exemption means the REC has not conducted a full ethical review, which would include an assessment of the technical soundness of the research methodology. This waiver should thus not be interpreted as a full ethical clearance. Rather, based on the considerations above, the REC sees the risks for the participants as minimal in relation to the social, educational, or scientific value of the research.

The Committee requests you to inform the Committee if substantive changes to the protocol are made. Moreover, the Committee requests you to send the

final report of the research containing a summary of the study's findings and conclusions to the Committee, for research managing and training purposes of the REC.

Wishing you all the best with your research,

Sandra Alba
Co-chair of the KIT REC

